Out of balance

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MOTHERING AFTER VERY PRETERM DELIVERY

Evaluation of the Infant Behavioral Assessment and Intervention Program on mother-infant interaction, maternal psychological distress, maternal parenting stress and maternal attachment representations

DOMINIQUE E. MEIJSSEN

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Dominique Emmelien Meijssen

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PROEFSCHRIFT

ter verkrijging van de graad van doctor aan de Universiteit van Tilburg, op gezag van de rector magnificus, prof. dr. Ph. Eijlander, in het openbaar te verdedigen ten overstaan van een door het college voor promoties aangewezen commissie in de aula van de Universiteit

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1

Introduction

'Het enige wat ik me van de bevalling herinner is dat het toch ineens moest gebeuren, ook al lag ik al drie weken in het ziekenhuis. Ik moest onder narcose. Ik werd wakker en toen kwam mijn vriend naar me toe, en die zei we hebben een zoon. Ik dacht wie ben jij, een zoon? De dag daarna ben ik pas naar T. gebracht. Bij het woord bevalling voel ik ook niets. Ik was ziek, moest geopereerd worden, er lag een patiënt in het ziekenhuis en dat was mijn zoon. Ik moest met mijn bed naar de NICU, en toen werd ik met mijn bed naar een van die glazen huisjes gereden en zeiden ze dat is uw zoon. Toen dacht ik nog, het had net zo goed een van die andere huisjes kunnen zijn. Ik kreeg allerlei scenario's in mijn hoofd van die enge films waarin kinderen worden verwisseld.'

(English translation: Appendix II)

INTRODUCTION

The birth of a baby is a major life event, involving psychological adjustment by both parents and changing family roles and relationships. Preterm birth, however, interrupts the normal process of adjustment to parenthood, not only because of the unexpected nature of the birth, but also because of the infant's fragile medical condition and extended hospitalization. Although most preterm babies survive, they are at increased risk of neurodevelopmental, social-emotional and health problems. These problems make mutually satisfying parenting and interaction difficult to achieve. In addition such problems may contribute to parental and parenting stress. Intervention programs focused on both infant development and parent-infant interaction may promote infant developmental outcome and mutually satisfying parenting.

Preterm birth, definitions and causes

Preterm birth is defined by gestational age as a degree of maturity. Three subgroups are distinguished by the World Health Organization (WHO, 2007): preterm (<37 weeks' gestation), very preterm (<32 weeks), and extremely preterm (<28 weeks). In the United States of America and several other countries a classification according to birth weight is generally used. Low-birth-weight infants are defined as those with a birth weight of 2,500 grams (g) or less, which may be due to prematurity, being born small for gestational age (SGA), or both. Similarly, lower cut-off limits for weight have been used to describe more severe cases, i.e., very low birth weight (VLBW <1,500 g) and extremely low birth weight (ELWB <1,000 g) (Sherry, Mei, Grummer-Strawn, & Dietz, 2003).

Preterm birth affects almost 12-13% of pregnancies in the United States and 5-9% in Europe and developed countries (Mercer et al., 2000). In the Netherlands preterm birth occurs in 7 to 8% of all the pregnancies (http://www.perinatreg.nl). Various risk factors have been consistently associated with preterm birth, such as multiple pregnancy, low socioeconomic status, second teenage pregnancy, parity and past reproductive history, substance abuse, infection and hypertensive disease during pregnancy (Slattery & Morrison, 2002). Approximately 14% of the variation in gestational age is explained by maternal genetic factors, and 11% by fetal genetic factors (Lunde, Melve, Gjessing, Skjaerven, & Irgens, 2007). The fetal and maternal genotypes modify the risk of preterm delivery. Environmental factors such as infection, stress, and obesity, which activate inflammatory pathways, have been associated with preterm birth, suggesting that

environmental and genetic risk factors might operate and interact through related pathways (Crider, Whitehead, & Buus, 2005).
Between 1983 and 2006 the amount of preterm births has more than doubled in the Netherlands (De Kleine et al., 2007). The most important factors contributing to this increase of preterm birth are a rise in multiple pregnancies due to fertility treatments, the higher age of pregnant women, earlier intervention in problem pregnancies, and a better registration of both gestational age and extreme preterm births at the border of viability (Dijkstra, 2002).

Preterm birth: consequences for the infant
Because of technological advances in neonatal care the survival rates of very preterm infants have increased. However, biologically, due to their immature organs preterm infants are more susceptible for long-term neurological and health problems than term infants (Saigal & Doyle, 2008). Common neonatal morbidities are bronchopulmonary dysplasia (BPD), retinopathy of prematurity, necrotizing enterocolitis, and infection. Preterm infants are also at increased risk for brain injury. Studies based on neuropathological analysis and conventional brain imaging of premature infants have provided evidence for injury to cerebral white matter (Volpe, 2001; Childs et al., 2001; Inder, Warfield, Wang, Huppi, & Volpe, 2005). The principal lesions include periventricular hemorrhagic infarction and periventricular leukomalacia. Neonatal morbidity as a result of pulmonary immaturity, intracranial events and infections in the preterm infant consequently have been associated with poor developmental outcome in infancy and at school age (Stephens & Vohr, 2009). Almost 10-17% of all very preterm infants are diagnosed with severe handicaps before the age of ten (Veen et al., 1991; Voss, Neubauer, Wachtendorf, Verhey, & Kattner, 2007). Around 50% of all very preterm infants develop disabilities or impairments at school age (Saigal et al., 2003). These impairments relate to attention, behavior, visual-motor integration, language performance and/or academic skills (Saigal et al., 2003; De Kleine et al., 2007; Verloove-Vanhorick & Verwey, 1997).
The normal process of brain maturation that occurs during the last trimester of pregnancy is interrupted by preterm birth (Volpe, 2001). Organizational processes during this period, also referred to as the wiring of the brain, are important for development of autonomic stability, motor maturity, state organization, attention, interaction and self-regulation (Lester & Miller-Loncar, 2000). This wiring of neuronal circuits is regulated by endogenous factors as well as by sensory input and experience (Penn & Shatz, 1999). The impact of the NICU environment on the long term development of the

brain has been acknowledged (Als et al., 2003). Genetics determine the basic architecture of the brain, but its final form is dictated by experiences in the environment. If an infant's or child's experiences are abnormal, non-nurturing, traumatic, and/or chronically stressful during a time of immense change in the development of the brain, these experiences may result in learning processes that may leave a permanent imprint on the structure and mechanisms of the functioning brain throughout life. In the NICU preterm infants can show marked changes in oxygen levels and cerebral blood flow during stressful procedures. When these occur in the first few days of life, they can have devastating long-term effects on the infant because they increase the risk of intraventricular hemorrhage and periventricular leukomalacia (Maroney, 2003). Therefore, attention in the NICU's has shifted towards neuroprotective care strategies and neurodevelopmental support, in order to improve developmental outcome (Wielenga et al., 2009).

Next to and due to their vulnerable brain and medical complications, preterm infants often experience difficulties in self-regulation, referring to the ability to modulate emotion, self-soothe, delay gratification, and tolerate change in the environment (DeGangi, 2000; Clark, Woodward, Horwood, & Moor, 2008; Wolf et al., 2002). Preterm infants are more difficult to bring to an attentive state (i.e., hypo-aroused) and once they are, they are more easily over- aroused, requiring more sensitive fine-tuning of stimulation to maintain their attention and positive affect (Als, 1983; Eckerman, Hsu, Molitor, Leung, & Goldstein, 1999). This may affect their ability to explore and interact with their social and object environment and consequently this may affect their learning processes.

Preterm birth: consequences for the parents

Coping with preterm birth can be a difficult and distressing experience for parents (O'Brien, Heron Asay, & McCluskey-Fawcett, 1999). Parents of young children with very low birth weight reported more financial, social, and family stress than parents of healthy term born children (Cronin, Shapiro, Casiro, & Cheang, 1995; Singer et al., 1999; Taylor, Klein, Minich, & Hack, 2001). The emotional adjustment of the mother may be compromised, resulting in negative consequences for the infant (Barry, Dunlap, Cotten, Lochman, & Wells, 2005; Murray et al., 1999). Mothers of preterm infants experience elevated levels of stress in the neonatal period (Singer, Davillier, Bruening, Hawkins, & Yamashita, 1996). They are more likely to suffer from depression and anxiety at the time of hospital discharge (Singer et al., 1996) and even during the first 18 months after birth (Kersting et al., 2004; Singer et al., 1999; Singer et al., 2003). Between

26 to 41% of the mothers of very preterm infants actually reported post traumatic stress symptoms (Kersting et al., 2004). Preterm birth not only has consequences for the emotional adjustment of the mother, but possibly also requires adjustment in parenting. Parenting a preterm infant can be more difficult than parenting a term infant because of their central nervous system immaturity, which places them at increased vulnerability for short- and long-term developmental problems. The altered behavioral responses and sleep-wake patterns that differ significantly from term infants (Holditch-Davis, Brandon, & Schwartz, 2003) complicate parenting issues and mutual interaction processes. Even basic parent-infant interactions such as feeding, handling, and engaging might be compromised (Minde, 2000). Study results are conflicting, some studies reported more parenting stress in mothers of high-risk very low birth weight infants (Auslander, Dvorah, & Arad, 2003; Drotar et al., 2006; Singer et al., 1999; Singer et al., 2007; Taylor et al., 2001). Other studies did not find an influence of infant medical risk status on parenting stress (Candelaria, O'Connel, & Teti, 2006; Saigal, Burrows, Stoskopf, Rosenbaum, & Streiner, 2000; Halpern, Brand, & Malone, 2001). Parenting stress has a negative effect on the parents' well being, and may also impair their ability to respond constructively to their child's needs (Deater-Deckard & Bulkley, 2000).

Preterm birth: consequences for the mother-infant interaction

Normal interaction moves from matching (synchronous) dyadic behavioral states to mismatching engagement states and communicated meaning with reparation back to matching states (Tronick & Cohn, 1989). To develop an interaction with effective and sufficient reparation of mismatches, maternal sensitivity and responsivity are important requiring accurate perception of cues and appropriate and timely responses. Especially the vulnerable preterm infant could profit from the experience of synchrony and the mutual ability to repair interactional mismatches as these are important foundations for infant growth and shape the child's cognitive, symbolic, self-regulatory, and social-emotional development (Feldman & Eidelman, 2004; Tronick, 2007). The dyadic system between parents and preterm infants, however, is less synchronous and mutually adaptive than in dyads with term infants (Feldman, 2006).

The preterm infant is known for its unclear behavioral signals due to neurological immaturity. In general preterm infants have been commonly characterized as being less actively involved in interaction; they look at their mothers less, show more gaze aversion, show less positive and more negative facial expressions and vocalize less (Eckerman et al., 1999; Goldberg & DiVitto, 1995; Tronick, Scanlon, & Scanlon, 1990).

Some studies have shown that maternal behavior mimics preterm infant behavior, resulting in less active and less responsive interactive behavior (Censullo, 1994). Other studies have shown the opposite, with mothers being more directive, more active in initiating and maintaining interactions and more stimulating throughout the first year (Crnic, Ragozin, Greenberg, Robinson, & Basham, 1983; Holditch-Davis, Schwartz, Black, & Scher, 2007). The repetitiveness of child care routines and the lack of reinforcement, which may accompany mothering a preterm infant, can become a source of maternal frustration. Mothers may become intrusive or even "aggressive" with their infants to compensate for perceived developmental and behavioral delays (Barnard et al., 1989; Swartz, 2005). However, a review on premature infant-mother interaction concluded that mothers of preterm infants do not seem to display as much intrusive or non-contingent behavior, as was seen in the past (Bozzette, 2007). Overall, mothers of preterm infants have to put more effort to initiate and maintain interactions and they also receive fewer positive responses from their infants, than mothers of full term infants (Singer et al., 2003).
Besides factors in the infant, such as illness severity, factors in the mother and her social-demographic background influence mother-infant interaction both after preterm and term delivery. Maternal depression is negatively related to the quality of mother-infant interaction, but positively to paternal support and a higher maternal educational level (Korja et al., 2008; Lee, Holditch-Davis, & Miles, 2007).

Preterm birth: consequences for the mother-infant relationship

The mother-child relationship is at risk after very preterm birth. The fragile emotional state (feelings of fear, guilt, and distance) of the mother may influence the way mothers relate to their baby. But also the NICU setup may create barriers to the natural growth of the mother-child relationship, because of difficulties with feeding and bonding (Davis & Stein, 2004). This relationship with the infant is very important for the parental motivation for interacting sensitively with the infant, supporting the infant's development, and to develop caregiving competencies. Not only in the NICU is the mother-child relationship at risk, but also at home after discharge. Parents often need to get to know their infant and its reactions to the new home environment. Due to self-regulatory difficulties in the infant, the infant may sleep little, cry a lot and have feeding difficulties. Breast feeding may be a struggle which may leave mothers again with feelings of failure and guilt, if it does not work out well for her and her baby. The fact that preterm babies often show negative affect and avoid eye contact (Eckerman et al., 1999) makes parenting and relating to the baby less rewarding than

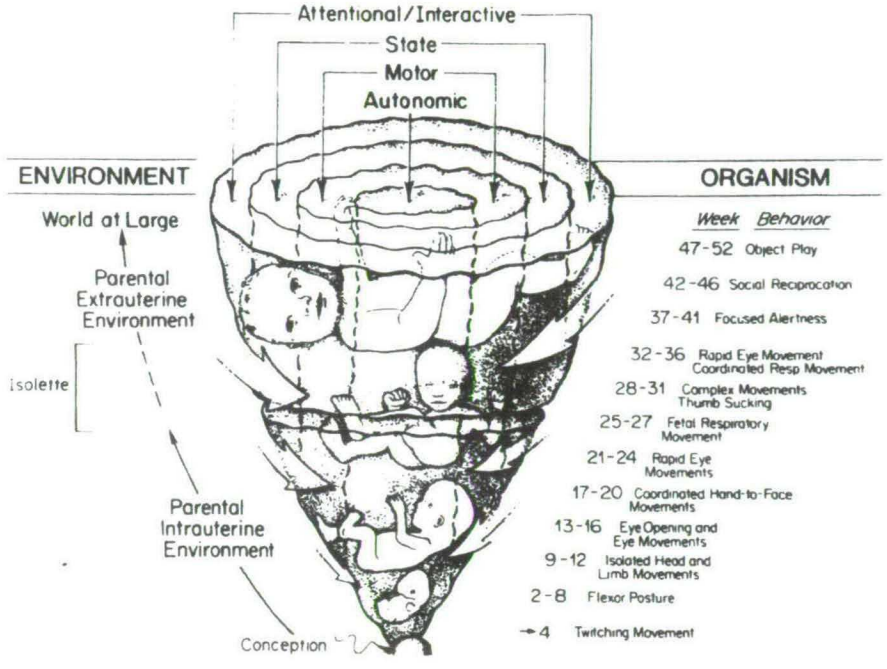
for parents of healthy term babies.
Despite the existence of some studies on the effect of preterm birth on infant attachment relationships, the results remain inconclusive. One study found more insecurely attached infants in a preterm group compared to a term group of infants (Mangelsdorf et al., 1996), an other study found comparable amounts of insecure and securely attached preterm and term infants (Brisch et al., 2005). Maternal mental representations of attachment and caregiving could serve as a risk or a protective factor, affecting parent-child interaction, the infant's motor, mental and emotional development and infant attachment (Brisch et al., 2005).

Intervention programs
Considering the fact that developmental, social-emotional and health problems of very preterm infants persist even into early adulthood (Saigal et al., 2008) , various types of early intervention programs have aimed to improve the long-term outcome of these children. Recent meta-analyses (Spittle, Orton, Doyle, & Boyd, 2007; Vanderveen, Bassler, Robertson, & Kirpalani, 2009) showed that post-discharge interventions that focus on the parent-infant relationship, along with infant development, have the greatest impact on cognitive development in the short to medium term. Because of the strong heterogeneity between the different intervention programs it is still unclear which aspects of early developmental interventions affect outcome the most. Questions still are: what is the optimal duration of the intervention, the best age to begin with the intervention, the optimal frequency and/or the focus of intervention.
Further research is needed to determine the components of intervention that are most effective, also taking into account benefits as well as costs. The above mentioned meta-analyses, however, have not investigated the effects of intervention programs on the child's behavior, parental outcomes (such as depression and anxiety), function, activity levels or participation, which may also be influenced by early developmental intervention programs.
Early self-regulatory support, as one of the basic elements underlying the infant's development and parent-infant relationship and interaction, may be an important target area for early intervention. As mentioned before, preterm infants experience difficulties in regulation of arousal and emotional responses and in flexibly allocating and sustaining their attention (Clark et al., 2008). Initially, the primary caregivers co-regulate the infant's postnatally developing central and autonomic nervous system (Schore & Schore, 2008). When the caregivers contingently tune their activity level to the infant during periods of social engagement, the infant is allowed to

recover quietly in periods of disengagement, and the more the caregiver attends to the infant's reinitiating cues for reengagement, the more synchronized the interaction becomes. The dyadic regulation of emotion is mediated by the attachment relationship.
As a result of neurophysiological development the infant's self regulatory capacities become gradually more efficient, needing less co-regulation from their environment. One post-discharge intervention program was specifically aimed at supporting the infant's self-regulatory competence (Kaaresen, Ronning, Ulvund, & Dahl, 2006). This intervention program is a modified version of the Mother-Infant Transaction Program (MITP). No effects were found on infant development, but the intervention resulted in less parenting stress at two years of age (Kaaresen et al., 2006; Kaaresen et al., 2008). A comprehensive post-discharge intervention program that focuses on sensitive parent-infant interaction and infant's self-regulation may be beneficial for both infant development and parental outcome, such as psychological well being and parenting experiences.

The Infant Behavioral Assessment and Intervention Program (IBAIP©)
In search of a comprehensive intervention program, the IBAIP showed promising results with regard to the neurobehavioral development and self regulatory competence of very preterm infants until 6 months of corrected age (Koldewijn et al., 2005). The IBAIP (Hedlund, 1998) is based on the synactive theory of behavioral development organization (Als, 1986). In this model (Figure 1) the infant's functioning is considered as handling experiences in continuous interaction with its environment.
The infant is described as a system consisting of four subsystems; the autonomic, motor, state, and attention/interaction system. The infant utilizes behaviors within each of the four subsystems to realize communicative or interactional goals. Three categories of behaviors have been identified: approach, self-regulatory, and stress behaviors. The autonomic system is behaviorally observable in the pattern of respiration, color changes, and visceral signals; the motor system is expressed through changes in tone, posture and movement; and the state subsystem is expressed via the range of states available for the infant, the clarity of any state, and patterns of transition from one state to the other, varying from deep sleep to crying. These subsystems allow the infant to: 1) engage in exploration and processing of cognitive and social-emotional information; 2) stabilize himself during this process of engagement, or defend himself by momentarily breaking the intensity of the social or cognitive interaction; or 3) remove himself from environmental and social stimuli by terminating the interaction. The subsystems are at all times interactive with the

Figure 1 Model of the synactive organization of behavioral development *[from Als H., 1982: with permission]*



current environment. The infant shapes his own environment by selecting information and by initiating and eliciting action in others. The environment, in turn, constantly provides opportunities and challenges either to be taken or avoided. If the level of input and information is appropriate for the infant, the infant may show approach behavior and take a step forward in development. Self-regulatory behaviors may be used by the infant to maintain balance or to return back to balance. They can support the infant in concentrating on stimuli that are offered to him or console the infant if the stimuli are overwhelming and inappropriate. Stress behaviors indicate that the sensory input is too intense, too frequent, too long or too complex for the infant. Although behaviors are categorized as approach, self-regulatory or stress, their interpretation may vary depending on the way they are used by the infant. Each behavior may be viewed as part of a continuum.

The goal of the IBAIP is to enhance the infant's social and environmental interactions without distress, reinforcing the infant's motivation and autonomy to explore and to learn from interactions. It is based on the assumption that the parent's availability and adequate responsiveness strengthens the infant's regulatory competence and development.

The intervention is guided by the Infant Behavioral Assessment (IBA, Appendix III) (Hedlund & Tatarka, 1988) and performed by specially trained professionals, hereafter referred to as interventionists. The IBA is an observational tool which helps the interventionist to make parents aware of their baby's responses to information. Based on these behavioral observations the interventionist assists parents to support their infant's self-regulatory efforts and to adjust the environment to match the neurobehavioral needs of the infant, like support of posture and a graded input of information. Monitoring the degree and quantity of input and support offered to the infant and when and under what circumstances they should be increased or decreased is of paramount importance in supporting the infant's self regulatory competence. The intervention is also based upon the conceptualization of the zone of proximal development (Vygotsky, 1978). The zone of proximal development refers to the distance between the actual developmental level as determined by independent problem solving, and the level of potential development as determined through problem solving under adult guidance or mediation. Within the infant's zone of proximal development there are degrees and quantities of neurobehavioral strategies that can be offered to the child. Input and support are maintained at the present level, increased or decreased, depending upon the infant's responses. The interventionist's ongoing assessment and scaffolding of the different neurobehavioral strategies, is critical to ensuring optimal infant interaction with the caregivers and the environment at large.

The intervention program consists of 6-8 home visits lasting approximately one hour. After each intervention session the parents receive a written report, illustrated with pictures of their infant. This report describes the infant's neurobehavioral and developmental progress and gives suggestions how to support the infant's explorations and self-regulatory competence. As the infants mature and their neurobehavioral functioning becomes increasingly stabilized, parents are encouraged to gradually reduce their co-regulatory support and to enjoy their infants' growing independency.

Central to the IBAIP is the support of the parent to raise their child. Mindful attention to their infant's behavioral expressions and development may enhance the parent-infant relationship, feelings of joy, and confidence in themselves and their child.

Aim of the study

In a pilot study the IBAIP was proven beneficial for the neurobehavioral development and self regulatory competence in very preterm infants until 6 months of corrected age (Koldewijn et al., 2005). This led to the

hypotheses that the IBAIP, next to long term developmental improvements in children, may also have positive effects for the parent, i.e., the parent-infant interaction, parental psychological well being, parenting stress and the parent-infant relationship. Therefore a randomized controlled trial was designed in which both child and parent outcome measures were used. In 2004 the inclusion of the families started and 176 infants and their parents were followed up to 2 years of corrected age (Figure 2).

The aim of the part of the study presented in this thesis, was to evaluate the effects of the IBAIP on: 1) mother-infant interaction; 2) parental psychological distress; 3) parenting stress; and 4) maternal attachment representations. We expected the IBAIP to promote positive and sensitive maternal interaction behavior, as sensitive interaction and joyful experiences are key components of the IBAIP. During the Still-face procedure, in which mother-infant interaction is interrupted by a 2-minute flat facial expression of the mother, we expected in the infants who received IBAIP to see more positive and explorative interaction behavior during interaction and better self-regulatory competence and fewer stress symptoms during the Still-face episode.

Parental psychological distress and parenting stress may be less after receiving IBAIP in which the parents are intensively supported in raising their child. The IBAIP might result in making the parents feel satisfied with and confident in themselves and their child leading to less psychological distress (e.g., anxiety) and parenting stress.

We also expected maternal attachment representations to be positively influenced by the IBAIP, resulting in more balanced representations, due to the attention for the mother-infant interaction processes during the IBAIP.

The assessments of the effects of the IBAIP and the consequences of very preterm birth for parents, varied from observational tools with micro-analytic coding systems, to in depth interviews and self-report measures.

Figure 2 Patient flow and follow up

Note:
GHQ; General Health Questionnaire (Chapter 3)
NOSIK; Nijmeegse Ouderlijke Stress Index Kort (Chapter 4)
NOSI; Nijmeegse Ouderlijke Stress Index (Chapter 4)
WMCI; Working Model of the Child Interview (Chapter 5)

Outline of the thesis

This thesis is divided into six chapters. Following the first introductory chapter (**Chapter 1**), the effect of the IBAIP on the mother-infant interaction is addressed in **Chapter 2**. During the Still-face procedure both mother and infant interaction behaviors are assessed and maternal sensitivity and responsivity is coded. In **Chapter 3** the effect of the IBAIP on maternal psychological well being (from 6 to 24 months after birth) is examined. **Chapter 4** describes the effect of the IBAIP on maternal parenting stress at 12 and 24 months after birth. In order to explore if the IBAIP affects maternal representations of the attachment to her child, **Chapter 5** reports on the results of the Working Model of the Child Interview. The WMCI is an in depth interview to give insight in the perceptions of the parent concerning the infant and the relationship with the infant. Finally, **Chapter 6** ends with a general discussion, followed by a critical review of the strengths and limitations of the study, the implications for prevention and intervention and considerations for future research. Throughout the thesis the chapters are illustrated with quotes from individual mothers and families.

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2

The effect of the Infant Behavioral Assessment and Intervention Program on mother-infant interaction after very preterm birth

'Na 2,5 maand kwam hij thuis, een periode waar ik me zo op had verheugd, om lekker samen te zijn. Maar dat gebeurde eigenlijk helemaal niet, omdat hij alleen maar aan het huilen was. Wat ik ook deed, en ik ging echt heel ver voor hem want ik had ook het gevoel dat ik iets in te halen had, maar het was alsof niets goed was…'

THE EFFECT OF THE INFANT BEHAVIORAL ASSESSMENT AND INTERVENTION PROGRAM ON MOTHER-INFANT INTERACTION AFTER VERY PRETERM BIRTH

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Abstract

Background: Prematurity and perinatal insults lead to increased developmental vulnerability. The home based Infant Behavioral Assessment and Intervention Program (IBAIP) was designed to improve development of preterm infants. In a multicenter randomized controlled trial the effect of IBAIP was studied on mother-infant interaction as a secondary outcome.

Method: Mother-infant interaction was assessed during the Still-face procedure at 6 months corrected age. 112 Mother-infant dyads (57 intervention, 55 control) were studied.

Results: Findings partially supported our hypothesis that the intervention would increase maternal sensitivity in interaction with their preterm infants. No effects were found on infant self-regulatory behavior or positive interaction behavior.

Conclusion: The family-centered and strength-based approach of IBAIP appears to be a promising intervention method to promote sensitive mother-infant interaction at home after discharge from the hospital. However, no positive effects were found on infant interaction behavior.

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INTRODUCTION

The early mother-infant relationship is at risk when an infant is born very preterm (< 32 weeks of gestation). Research on brain development has confirmed the importance of the interaction between the environmental and social stimulation and neurobiological processes for the development of brain structures (Smith & Thelen, 2003; Gunnar & Quevedo, 2007; Friedman, 2006; Schore, 1994). For preterm infants 'normal' environmental stimulation can cause over-arousal that may result in serious distress, which might influence brain development in a negative way (Als et al., 2004; Graham, Heim, Goodman, Miller, & Nemeroff, 1999). Postnatally preterm infants also have more difficulties interacting with their environment, including their parents (Swartz, 2005). They initiate interaction less frequently, show less positive affect, and are less attentive (Belsky, 1984; Goldberg & DiVitto, 1995; Tronick, Scanlon, & Scanlon, 1990). They are difficult to bring into an attentive state, and once in that attentive state, they are easily over-aroused, show more negative affect and often avoid eye contact (Eckerman, Hsu, Molitor, Leung, & Goldstein, 1999).

For parents, their preterm infant's behavior may result in greater difficulty in interaction than with a term infant. Consequently, preterm infants may not be stimulated or responded to in an optimal way. Some studies have shown that parenting behavior mimics preterm infant behavior, resulting in less active and less responsive interactive behavior (Censullo, 1994). Other studies have shown the opposite, with parents being more directive, more active in initiating and maintaining interactions and more stimulating throughout the first year (Crnic, Ragozin, Greenberg, Robinson, & Basham, 1983; Holditch-Davis, Schwartz, Black, & Scher, 2007). The repetitiveness of child care routines and the lack of reinforcement, which may accompany parenting a preterm infant can become a source of parental frustration. Mothers may become intrusive or "aggressive" with their infants to compensate for perceived developmental and behavioral delays (Barnard et al., 1989; Swartz, 2005). However, a recent review on premature infant-mother interaction concluded that mothers of preterm infants do not seem to display as much intrusive or noncontingent behavior, as seen in previous studies (Bozzette, 2007).

Preventive early (post-hospital discharge) interventions directed at the interaction between preterm infants and their parents to improve developmental outcome have been promoted in several studies (Spittle, Orton, Doyle, & Boyd, 2007; Bonnier, 2008). The largest effects were found on improving cognitive development in the short to medium term. In these programs special attention was also paid to the individual infant's

capacities and responses to stimulation, as well as to the individual parent's way of handling, initiating contact and responding to their child. Few intervention studies address the infant's self-regulatory competence during early interactions. Self-regulatory competence is important for the infant's social interactive and exploratory opportunities, which are necessary for learning processes (Bronson, 2000). The Infant Behavioral Assessment and Intervention Program is an intervention program that guides the parents in supporting their infants' self-regulatory competence (IBAIP) (Hedlund, 1998).

In previous work, we have shown in a randomized controlled trial carried out in Amsterdam, the Netherlands, that the IBAIP enhances the mental, motor and neurobehavioral outcome of very preterm infants (<32 weeks and/or <1500 grams) at 6 and 24 months corrected age (Koldewijn et al., 2009). In this paper we focus on the mechanisms that may have contributed to the developmental improvements in the children. The mothers in the intervention group were provided detailed information regarding their infants' behavior and encouraged to show responsive and engaged behavior towards their infants. Therefore, the effect of the IBAIP on mother-infant interaction after the intervention period (6 months corrected age) was studied, using the Still-face procedure (Tronick, Als, Adamson, Wise, & Brazelton, 1978). The Still-face procedure consists of three short episodes: one of normal mother-infant play interaction, followed by a mildly stressful period when the mother does not interact with the infant and keeps a blank face, and followed by an episode when the mother resumes the playful interaction. We hypothesized that the IBAIP would: 1) improve self-regulatory competence of the infants expressed in a different pattern of behaviors across the three episodes, with better coping responses and fewer stress symptoms in the Still-face episode or more positive and explorative interaction behavior during the interaction episodes; 2) promote maternal positive and sensitive interaction behavior throughout the interaction in both the first and the third episode.

METHOD

Participants

Two hospitals with level III NICUs and five city hospitals participated in this trial. All infants with a gestational age of <32 weeks and/or a birth weight of <1500 grams, admitted to one of these seven hospitals and whose parents were living in Amsterdam, were eligible for the study if they survived to a post menstrual age of 32-34 weeks. Infants with severe congenital abnormalities, infants whose mothers had a documented history of illicit

drug use or severe physical or mental illness, infants from non-Dutch speaking families for whom we could not arrange an interpreter, and infants who participated in other trials on post discharge management were excluded from the study. At 35-38 weeks post menstrual age, the participating infants were randomly assigned to the intervention group or the control group. The randomization was computer-generated at the Academic Medical Center and stratified for gestational age (<30 and ≥30 weeks) and recruitment site; twins were always assigned to the same group.

Recruitment took place from January 2004 to April 2006; 315 infants were eligible for the study. The parents of 38 infants refused participation, 11 infants died before recruitment, 38 infants were excluded and 52 participated in another post-discharge trial. Finally, 176 infants were available for randomization (151 families); 86 infants and their 72 parents were assigned to the intervention group and 90 infants and their 79 parents to the control group (for a detailed description see Koldewijn et al., 2009). Data from 112 mother-infant dyads was available for analysis, 57 in the intervention group and 55 in the control group. Data was lost because of the absence of the mother and the presence of the father (n=12), language problems (n=7), infant's age at recording (> 8 months corrected age, n=1) or too many interruptions during the procedure (n=4). Some data could not be gathered because of parents refusing participation (n=2), infants' illness (n=3), infant's death (n=1), no-shows (n=3), or technical problems (i.e., recording difficulties with video or split-screen apparatus, n=31).

Maternal sensitivity and responsivity was analyzed in 109 mothers-infant dyads, 53 in the intervention group and 56 in the control group. Data of 3 mothers could not be coded as mothers' hands could not be seen, a specific condition for coding the Maternal Responsivity and Sensitivity Scales (MSRS).

Chi square tests and analyses of variance revealed no significant differences in neonatal and socio-demographic characteristics between the mother-infant dyads who were analysed and those who were not. Table 1 shows the neonatal and socio-demographic backgrounds of the 112 infants and their parents in both groups. No significant differences were found between the intervention and the control group with respect to parental background and neonatal characteristics.

Table 1 Neonatal and socio-demographic characteristics of intervention and control group

	Intervention (n=57)	Control (n=55)	P
Neonatal characteristics			
Gestation (wk), mean (SD),	29.5 (2.1)	30.1 (1.9)	0.08
range	(26-35)	(25-35)	
Birth Weight (g), mean (SD),	1246 (356)	1328 (335)	0.21
range	(640-2005)	(495-2095)	
Gender infant: male/female	33/24	25/30	0.19
Multiplets, n (%)*	20 (35%)	23 (42%)	0.09
Artificial ventilation n (%)	25 (45%)	19 (35%)	0.31
Intraventricular haemorrhage (IVH) n (%)▫	11 (22%)	8 (14%)	0.35
IVH grade I +II / III + IV	8 / 3	7 / 1	0.43
Periventricular leukomalacia (PVL) n (%)▫	8 (15%)	6 (11%)	0.54
PVL grade 1 / 2+3	7 / 1	5 / 1	0.82
Socio-demographic characteristics			
Maternal age, mean (SD), y	32.7 (5.3)	31.6 (5.2)	0.25
Paternal age, mean (SD), y	36.7 (7.7)	35.8 (6.1)	0.50
Firstborn child (%)	35 (64%)	27 (52%)	0.22
Country of birth, mother (%)			
Netherlands	33 (58%)	31 (57%)	
Surinam	10 (18%)	6 (11%)	
Morocco	4 (7%)	3 (7%)	
Other	10 (17%)	15 (25%)	0.33
Mother speaking Dutch	51 (89%)	42 (78%)	0.09
Maternal education (%):			
No high school graduate	20 (35%)	20 (36%)	
High school graduate	37 (65%)	34 (63%)	0.83

Analyses of variance and Chi-square test
Note: Numbers are given as number of infants unless otherwise stated
** at least 2 infants survived*
▫ Intraventricular haemorrhage (IVH) was defined according to Papile, Munsick-Bruno, & Schaefer (1983), ▫ Periventricular leukomalacia (PVL) according to De Vries, Eken & Dubowitz (1992)

Intervention

The IBAIP is a post-discharge preventive intervention program for infants up to the developmental age of 6-8 months and their parents (http://www.ibaip.org). It is based upon the conceptual framework of Heidelise Als (1986) that underlies the Newborn Individual Developmental Care and Assessment Program (NIDCAP; Als, 1986).The organization of the intervention is guided by the Infant Behavioral Assessment (IBA; Hedlund & Tatarka, 1988). The goal of the IBAIP is to enhance the infant's social and environmental interactions without distress and reinforce the infant's motivation and autonomy to explore and to learn. The IBA is an observational tool which helps the interventionist to sensitize parents to their baby's responses to information in order to assist parents in supporting their infant's self-regulatory efforts and to adjust the environment to match the neurobehavioral needs of the infant. It is based on the assumption that the parent's availability and adequate responsiveness strengthens the infant's regulatory competence and development.

The infants in the intervention group received 6-8 home visits by the interventionists, experienced pediatric physical therapists trained in the IBAIP, as well as standard care. To a certain extent, the frequency was tailored to the needs of the family (6-8). In practice only 2 families had 6 interventions, and 2 had 7, all others had 8. Standard care consisted of regular visits at the outpatient local pediatric clinic. After each intervention session (approximately one hour) the parents received a report, illustrated with pictures of their infant. This report described the infant's neurobehavioral and developmental progress and gave suggestions how to support the infant's explorations and self-regulatory competence. As the infants matured and their neurobehavioral functioning became increasingly stabilized, parents were encouraged to gradually reduce their co-regulatory support and to enjoy their infants' growing independence. The infants in the control group received standard care.

Instruments

The Still-face procedure follows a systematic protocol to observe the parent-infant interaction and code it with the Infant and Caregiver Engagement Phases (ICEP) (Weinberg & Tronick, 1999) and the Maternal Sensitivity and Responsivity Scales (MSRS) (Cenciotti, Tronick, & Reck, 2004). This procedure confronts the infant with an age-appropriate developmental task (face-to-face social interaction with the mother), an age-appropriate episode of mild stress (the mother holding a still-face and remaining unresponsive), and a reunion episode during which the infant and mother renegotiate the interaction after it has been disrupted by the

still-face (reunion face-to-face interaction with the mother) (Tronick, 2007). The mother sits in front of the infant at eye level and the infant is in an infant seat. The procedure starts with the face-to-face social interaction episode of 2 minutes in which the parent is instructed to interact with the infant. The parent can touch the infant, talk to the infant, but cannot use any toys in the interaction, except for objects inherent to the face-to-face setting. After these two minutes the parent turns around with her/his back to the infant for fifteen seconds. Then the parent turns back and the Still-face episode begins. The parent is not allowed to make any contact whatsoever and is instructed to look at the side of the infant seat. After the two minutes of Still-face the parent turns around again for fifteen seconds and then the reunion episode begins, in which the parent interacts with the infant for two minutes.
The Still-face was carried out in the hospital or in a home visit. In both settings only the researcher was present. Distracting stimuli (e.g., television or music) were removed to control for potentially confounding effects. The procedure was recorded with two cameras, one recording the parent and one the infant. The two images were combined using a split-screen module. For scoring the Observer 5.0 (Noldus) program was used. In this program all behaviors are scored at a micro-analytic level on a second by second basis using the ICEP coding system. The MSRS were used to assess the attunement of the mother to her infant during interaction.

Infant behavior

The ICEP coding system discriminates several different infant behavioral phases: negative engagement (negative facial expressions and/or whimpering, complaining, fussy, or crying vocalizations, protest, withdrawn) object/environment engagement, social monitoring or mother focused behavior (the infant's attention is directed towards the caregiver with a neutral or interested facial expression), social positive engagement (the infant displays facial expressions of joy particularly smiles, but occasionally coo and play faces), and sleep. The behaviors are mutually exclusive. Additional infant codes are oral self-comforting (the infant uses his body to provide self-stimulation), self-clasp (both infant's hands are touching), distancing (increasing the physical distance from the caregiver without engaging an object), and infant autonomic stress indicators (e.g., spitting up or hiccupping). Self-comforting and self-clasp behaviors have been described as reflecting infant attempts to manage distress during the Still-face phase of the procedure (Field, 1994). The expected infant reactions to the Still-face procedure (the 'Still-face effect') are increased gaze aversion and decreased positive affect during the Still-face episode and

a carry-over effect of these reactions into the reunion episode (Adamson & Frick, 2003; Field, 1994).

Caregiver behavior

The ICEP discriminates different caregiver behavioral phases: negative engagement (angry, hostile, stern, sad, sober or expressionless), hostile/intrusive, withdrawn, non-infant focused engagement, social monitoring without speech or with neutral speech, social monitoring with positive speech, social positive engagement (smiling), and exaggerated positive engagement (exaggerated laughter, play, surprise, mock and/or coo faces). These behaviors are mutually exclusive. Additional caregiver codes are rough touches and violations of the procedure, like touching the infant during the Still-face episode.

Infants' and mothers' behaviors were coded by a trained coder (DM) from DVD. The interrater reliability after training was 0.80 (index of concordance). Behavior of mothers and infants was coded in separate runs, as coding was done on several mothers or on several infants in random order. The coder was blind to group membership. Whenever a behavior was noted, the time at which the behavior began and ended, was recorded. The duration data resulted in a total percentage of time that the infant or caregiver was engaged in a behavior. The percentages for each behavior were computed for the separate episodes (normal play, still-face, reunion).

The MSRS were used to capture the attunement of the mother to the infant during interaction. The MSRS consist of three rating scales, the sensitivity/responsivity scale, the undercontrol/withdrawal scale, and the overcontrol/intrusivity scale. Sensitive and responsive maternal behavior is defined as consistent, contingent and appropriate reactions to the infant's behavior. Undercontrolling and withdrawal behavior is characterized by disengagement and lack of investment in the interaction, which often makes the interaction seem flat and boring. Overcontrolling and intrusive behavior is characterized by overstimulation, interruption, and noncontingent behavior. Overcontrolling mothers impose their own behavior without looking at the infant's reaction and behavior. The scales range from one for no visible signs of the behavior to five for consistent and strong signs of the behavior (e.g., persistent and strong overcontrol). The MSRS were coded by a trained coder (DM) from DVD. Pearson's correlation coefficients for interrater reliability were 0.77 for sensitivity, 0.90 for undercontrol and 0.70 for overcontrol after training.

Procedure

At the (corrected) age of six months the infants and their parents were invited to come to the hospital for an extensive assessment consisting of physical examination, IBA and Bayley Scales of Infant Development assessment and the Still-face procedure. If parents were unable to come to the hospital, they were visited at home, which was done in 21 (37%) intervention and in 16 (29%) control cases, a non significant difference. After a short introductory conversation first the Still-face procedure was carried out.

All assessors including the standard care providers were blinded to the infant's group assignment. In this single blind randomized controlled trial only parents and interventionists were aware of the group assignment. Parents were instructed not to inform the outcome-assessors.

The Medical Ethics Committee of all hospitals involved in this trial approved the study. This trial is registered with controlled-trials.com, number ISRCTN65502576. All participating parents signed informed consent.

Statistical analyses

Characteristics of the intervention and control group were compared with analyses of variance or Chi square tests. The data on the Still-face procedure were studied in two ways. First, analyses of variance for repeated measures were performed to analyze group differences in interaction behavior (ICEP) across the three episodes (Play, SF, Reunion). Group (intervention, control), infant gender (male, female) and place of recording (home, hospital) were used as fixed factors. Gender was chosen as a factor, because preterm boys have frequently been reported to show increased vulnerability and gender differences have been found during the Still-face procedure with full term infants (Weinberg, Tronick, Cohn, & Olson, 1998; Belsky, 1984; Tronick & Cohn, 1989). Place of recording might be important as recording in the strange hospital situation might be more stressful than at home.

Second, differences per episode of the Still-face procedure were studied with both multivariate and univariate analyses of variance, again with group, gender and place of recording as factors, as group differences could appear more clearly in one of the episodes or in one specific behavior.

Differences in the assessment of maternal interaction quality (MSRS) were studied with multivariate as well as univariate analyses of variance, as group differences could appear in the overall assessment, as well as only in specific aspects of behavior. Again group, gender and place of

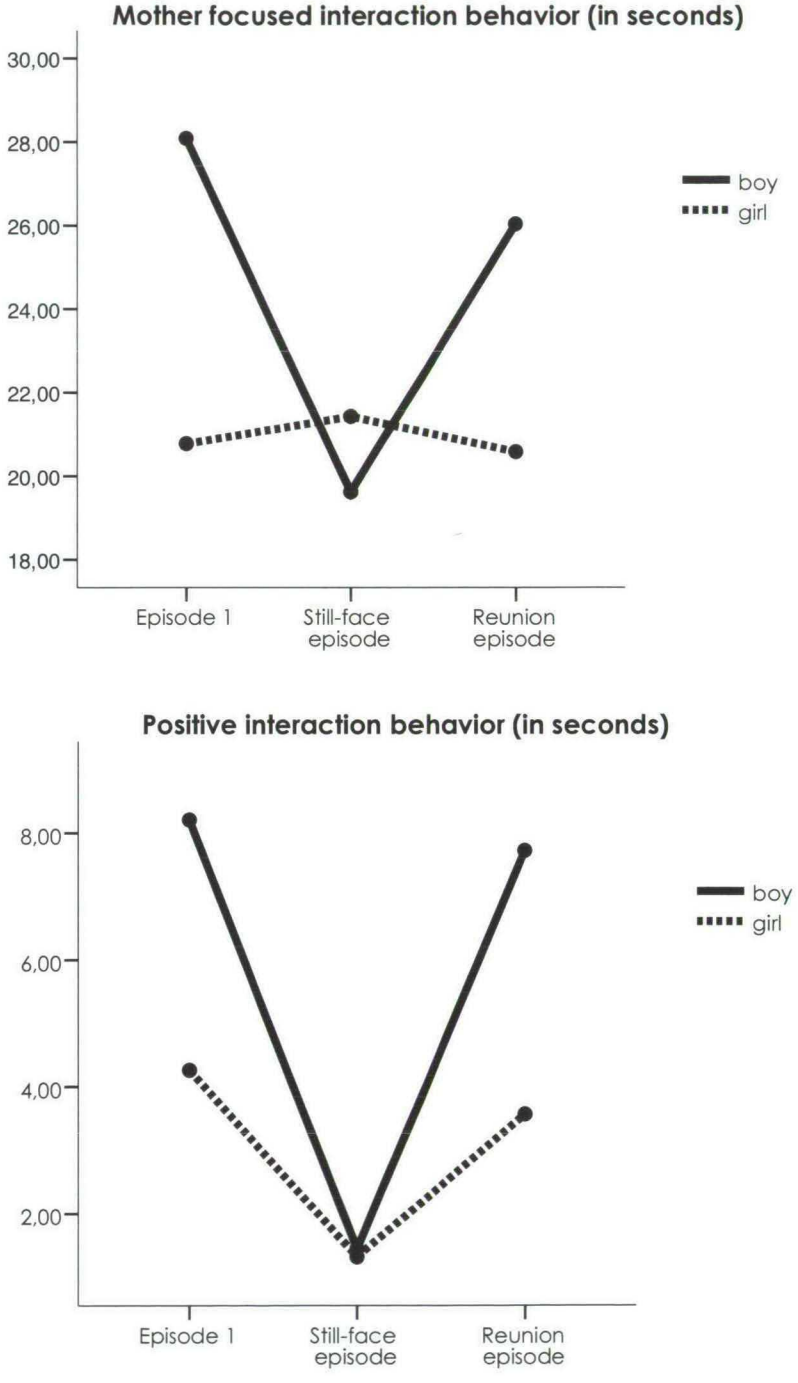
recording were used as factors.
Mother-infant interactions in mothers of twins were coded independently (one with each child). Assuming that mother-infant interaction is a process of mutual regulation, every mother-infant pair should be treated as independent. Twins were included together in the same group and the amount of twins did not differ between the intervention and control group. All statistical analyses were performed using version 14.0 of the Statistical Package for Social Sciences (SPSS 14.0, Chicago, IL, USA). An alpha level of 0.05 was used for tests of significance.

RESULTS

Preliminary analysis

First we evaluated whether gender or place of recording showed significant differences in behavior of the infants and mothers regardless of group membership. Gender differences were found for object/environment focused behavior, mother focused behavior and positive interaction behavior: boys looked more at their mothers than girls (mean difference=6.38, se=2.83, p=.03) and smiled more (mean difference=4.05, se=1.97, p=.04) (Figure 1), whereas girls were more object/environment oriented (mean difference=9.65, se=3.97, p=.02) during the interaction episodes (normal play and reunion episodes). Place of recording was also important for the infants' behavior. Neutral mother focused behavior was observed more frequently in the hospital (mean difference=8.08, se=2.67, p=.00) and object/environment focused behavior was observed more frequently in the recordings made at home (mean difference=8.86, se=3.77, p=.02).
These factors were incorporated into the analysis of the comparison of the intervention and control groups. No differences for gender or place were found for maternal behavior.

Figure 1 Gender differences during the Still-face procedure



Infant interaction behavior

The means and standard deviations of the percentage of time of the infants' interaction behavior are presented in Table 2. Distancing behavior or sleep was not observed and was not analyzed. The results of the analysis for repeated measures are reported for group, gender, place of recording, episodes and the group by episode interaction in Table 2 as well. For positive engagement a main effect for group ($p=.04$) was found, as well as an effect of episodes ($p<.01$) and an interaction effect for group by episodes ($p=.04$). The infants in the intervention group showed less positive engagement (smiling), especially during the first episode. For environment focused behavior a group by episodes interaction effect ($p=.02$) was found. Infants in the intervention group showed more environment focused behavior during the first episode (normal play) than the control group. No other between-group differences or group by episode effects were found. Our hypothesis that the infants in the intervention group would show more positive and exploratory behavior during the interaction episodes of the Still-face procedure was not confirmed.

No main effect for gender was found in these repeated measures analyses. Place of recording showed a significant main effect for both mother-focused behavior ($F(1,96)=8.45$, $p<.01$) and environment focused behavior ($F(1,96)=5.08$, $p=.03$). In the hospital the infants showed more mother-focused behavior and less environment focused behavior than in the home situation.

The Still-face effect is seen for both groups in the significant impact of episode on positive engagement, environment focused behavior, negative behavior and self-clasp behavior. The children showed less positive engagement and more environment focus and self-clasp behavior during the Still-face episode, and more negative behavior in the reunion episode. No differences were found in self-regulatory behavior or stress reactions of the infants throughout the procedure or in specific episodes related to group, hence our hypothesis was not confirmed.

Multivariate analyses on the first episode showed an interaction effect for group by place of recording ($F(7,98)=2.22$, $p=.04$). The infants in the intervention group focused less on the environment ($F(1,104)=$, $p=.02$) and showed more positive behavior (smiling) ($F(1,104)=$, $p<.01$) during the hospital recordings than the control infants. A main effect was found for gender ($F(7,98)=2.59$, $p<.01$) and place of recording ($F(7,98)=2.40$, $p=.03$), boys were less environment focused than girls $F(1,104)=11.34$, $p<.01$) and the infants recorded at the hospital were more focused at their mothers than those at home ($F(1,104)=9.35$, $p<.01$).

For the Still-face episode the MANOVA showed that place of recording

Table 2 Infant interaction behavior in % of time

Infants N=112	Normal play M (SD)		Still-face M (SD)		Reunion M (SD)		Group[1]	Gender	Place of recording	Episodes	Group x Episodes
Behaviors	Intervention	Control	Intervention	Control	Intervention	Control	F	F	F	F	F
Positive, smiles	4.7 (6.3)	8.1 (14.0)**	1.1 (2.4)	1.6 (4.3)	4.7 (6.8)	7.1 (14.4)	4.46*	3.38	1.09	18.99**	3.34*
Mother-focused	24.4 (16.8)	25.2 (14.6)	22.2 (17.0)	17.7 (17.2)	24.0 (18.0)	23.1 (17.1)	0.72	2.25	8.45**	1.61	0.68
Environment focused	66.5 (23.0)	63.4 (20.5)*	71.5 (21.7)	74.9 (24.0)	61.7 (24.8)	58.8 (26.0)	0.07	3.39	5.08*	21.74**	3.75*
Negative	2.8 (9.1)	1.6 (5.2)	4.7 (15.2)	3.2 (10.2)	9.3 (22.0)	9.0 (20.6)	0.16	0.13	1.27	10.55**	0.13
Stress	0	0.02 (0.14)	0.2 (1.2)	0.3 (1.5)	0.4 (2.0)	0.1 (0.6)	0.18	0.64	1.96	1.70	0.54
Oral S-C[2]	5.4 (13.0)	5.6 (10.5)	9.8 (14.9)	10.3 (17.0)	6.9 (11.9)	9.2 (16.3)	0.04	0.51	1.55	2.21	0.18
Self-clasp	2.3 (4.7)	1.2 (3.0)	2.5 (6.6)	3.9 (6.8)	1.0 (2.2)	1.1 (3.0)	0.07	0.38	0.59	3.33*	1.30

Note: Analyses of variance for repeated measures (corrected for infants gender and place of recording)
[1]Group: intervention or control (intervention effect); [2]Oral self-comforting: hand or finger touches mouth
**p≤.05, **p≤.01*

differed F(7,94) 2.91, p<.01). Children recorded at the hospital focused less at the environment F(1,104)=6.28, p=.014) and showed more oral self comforting F(1,104)=5.38, p=.02).
For the reunion episode the MANOVA showed no significant effects.

Maternal interaction behavior

Exaggerated positive engagement, rough touches and violations of the procedure were not observed. The mothers mostly showed neutral (62%) and positive social monitoring behavior (27%) and little negative behavior (0.08%). Table 3 presents the means and standard deviations of the percentage of time that the mothers showed the coded behaviors in the first play episode and in the last reunion episode. The results of the analysis for repeated measures are reported for group, episodes and the group by episode interaction in Table 3 as well.
An effect of episodes was found for neutral social monitoring behavior; mothers showed more neutral social monitoring behavior in the reunion episode. No main effects for group, gender and place of recording were found.
In the first play episode univariate analyses showed more positive behavior in the mothers in the intervention group (F (1,100) 3.92, p=.05). Multivariate analyses however, indicated no main or interaction effects in overall group, gender or place of recording in maternal behavior during the first play episode, neither during the reunion episode. Our hypothesis concerning more positive engaged interaction behavior of the mothers in the intervention group was not confirmed.

Table 3 Maternal interaction behavior in % of time

Mothers N=112	Normal play		Reunion		Group	Episodes	Group x Episodes
	M (SD)		M (SD)				
Behaviors	Intervention	Control	Intervention	Control	F	F	F
Negative	0.1 (0.67)	0 (0)	0.1 (0.51)	0 (0)	0.02	0.02	0.66
Non-infant focused	0.2 (0.73)	0.2 (0.69)	0.2 (0.78)	0.2 (1.1)	0.45	0.16	0.14
Social monitor/nvc	58.2 (20.1)	54.9 (23.7)	68.3 (25.5)	67.6 (25.3)	0.10	27.73**	0.70
Social monitor/pvc	24.5 (20.4)	28.6 (24.6)	27.2 (25.1)	28.2 (25.3)	0.33	0.86	1.05
Social positive engagement	3.9 (4.0)	2.5 (3.0)	3.5 (3.7)	2.7 (3.2)	3.5	1.58	0.33

Note: Analyses of variance for repeated measures (corrected for infants gender and place of recording)
nvc = no/neutral vocalisations, pvc = positive vocalisations
**p≤.05, **p≤.01*

Maternal sensitivity and responsivity

In Table 4 the results for the rating of maternal sensitivity and responsivity throughout the Still-face procedure are presented. Multivariate analyses of variance showed an interaction effect for group by gender ($F(3,98)=2.71$, $p=.05$), but none for the univariate comparisons of group by gender differed significantly. No other main or interaction effects were found on group, gender or place of recording. Univariate comparisons indicated that mothers in the intervention group showed more sensitivity and used less overcontrolling behaviors than the mothers in the control group (Table 4). This finding confirms our hypothesis.

Table 4 Maternal Sensitivity and Responsivity Scales (MSRS)

MSRS	Intervention group (n=53) M (SD)	Control group (n=56) M (SD)	F	DF	P
Sensitivity	4.13 (0.78)	3.91 (0.84)	3.80	1, 108	0.05
Overcontrol/Intrusiveness	1.75 (0.87)	2.04 (0.93)	4.29	1, 108	0.04
Undercontrol/Withdrawn	1.32 (0.55)	1.38 (0.68)	0.30	1, 108	0.58

Note: Analyses of variance (corrected for infants gender and place of recording)

Relationships between the micro-analytic ICEP and the more qualitative and general MSRS codes were explored using correlational analyses. Maternal positive engagement behavior was found to show significant correlations with maternal sensitivity in both interaction episodes. Moderately strong correlations were found for social monitoring behavior with or without positive vocalisations with the assessments of less over- and undercontrol (see Table 5).

Table 5 Bivariate correlations between MSRS scales and maternal interaction behavior during interaction episodes (normal play and reunion)

	MSRS		
Maternal behavior	Sensitivity	Overcontrol	Undercontrol
Episode 1 (Normal play)			
Negative	-.12	.01	.10
Non-infant focused	.01	-.09	.09
Social monitor/neutral vocalisations	-.03	-.12	.47**
Social monitor/positive vocalisations	-.02	.16	-.47**
Social positive engagement	.23*	-.26**	.02
Episode 3 (Reunion)			
Negative	-.12	.12	.11
Non-infant focused	.03	-.04	-.08
Social monitor/neutral vocalisations	-.04	-.16	.44**
Social monitor/positive vocalisations	.02	.19	-.43**
Social positive engagement	.25*	-.21*	-.09

Note: N=109
**p≤.05, **p≤.01*

DISCUSSION

The comparison of the intervention group and control group during interaction in the Still-face procedure showed a few subtle differences. The rating of the mothers' attunement in the intervention group was somewhat more sensitive and less intrusive than in the control group, hence our hypothesis regarding maternal behavior was partially confirmed.
The micro-analytic observation only showed a small difference in positive engagement behavior of the mothers. In contradiction to our hypothesis, the infants in the intervention group were found to show less positive behavior than the control infants and they focussed more on the environment, specifically during the first play episode. These behaviors may

be related. During exploration of their environment, smiling is less expected than in interaction with the mother. Interestingly, also an interaction effect of group by place of recording was found concerning positive behavior and exploration of the environment. During the recording in the potentially more stressful hospital environment, the intervention infants smiled more and explored less than the control infants, whereas this was reversed in the recordings made at home. In both groups the children focused more on their mothers during the hospital recordings. Perhaps this shows context dependent functioning.
The increase in positive engagement behavior of the mothers could also be seen as more intrusive or as compensating for a less engaged infant. However this possibility seems less plausible, as positive engagement behavior of the mothers was associated with more sensitive maternal interaction behavior and less overcontrolling behavior.
The findings also suggest an alternative explanation about the decrease in smiling in the intervention group. At about 6 months of age term infants become increasingly interested in their environment (Legerstee, Pomerleau, Malcuit, & Feider, 1987) and in objects (Cohn & Tronick, 1987). Thus it might be that the intervention infants showed improved development in this behavioral pattern; that is the increase in maternal sensitivity of the intervention mothers fostered a normal developmental transition from a focus on the mother to a focus on objects. This observation also supports the previous finding that the infants receiving intervention compared to the non-intervention infants at 6 months of age were more advanced on the Bayley Scales of Infant Development (Koldewijn et al., 2009).

The hypothesis that the intervention group infants would show better regulatory behavior, for instance an easier repair of interaction during the reunion episode or fewer stress responses throughout the Still-face procedure or in specifically stressful episodes, was not confirmed. However, the ICEP does not allow a measurement of self-regulatory behavior in great detail. For instance, a distinction in infant's gaze aversion as either self regulatory behavior following emotional arousal, or as real object or environment focused behavior could not be coded. Further study should be done to observe more closely self regulatory behaviors of the infants.

Gender differences in the infants' reactions were found. Boys looked at their mothers more and smiled more during the interaction episodes than girls. These differences correspond to differences seen in several studies of the Still-face procedure (Tronick et al., 1989; Weinberg et al., 1998), in which infant boys displayed more positive as well as negative affect, focused

more on the mother, and displayed more signals expressing escape and distress, and demands for contact, than girls. Girls showed more interest in objects.

Along with the studies by Segal et al. (1995), Montirosso, Borgatti, Trojan, Zanini & Tronick (2008), Hsu & Jeng (2008) and Erickson & Lowe (2008) this is one of the few studies to demonstrate the sensitivity of preterm infants to the Still-face procedure. Hsu & Jeng (2008) compared Taiwanese 2 months old preterm (gestation range 24-34 weeks) and term infants during the Still-face procedure and they found longer duration of negative affect in preterm infants and shorter latency to negative states. In a sample of very low birth weight infants it was found that maternal responsiveness was strongly associated with the amount of positive infant affect, but not with negative infant affect (Erickson & Lowe, 2008). Erickson and Lowe (2008) suggested a relative chronicity or maintenance of negative affect and proposed that early relationship-focused intervention might increase parental responsiveness. A recent review of the Still-face paradigm showed that maternal interaction behavior is not necessarily associated with infant interaction behavior during the three episodes (Mesman, Van IJzendoorn, & Bakermans-Kranenburg, 2009). This fits our results: in the intervention group the mothers showed more sensitive and positive interactive behavior, but the infants did not show more positive interaction behavior in the intervention group.

Strenghts and limitations

A strength of our study is that it was a randomized control trial, however, a pretest-posttest design might more readily have found a significant improvement in interactive behavior. A design with a dummy-treated control group that controlled for potential attention effects of the intervention would have also been useful. Consequently, we cannot determine which part of the intervention procedure (only attention, or the actual information and support provided) is responsible for any of the results. Another potential limitation of the study is that it was done in a sample of mothers of preterm infants without a history of drug use or mental health problems, which limits the generalizability of the results to that population.

Usually the Still-face procedure is used in a laboratory setting, which was not feasible for us. The hospital setting reflected the laboratory setting better than the home setting, so we controlled for setting. In view of our results regarding smiling and exploration of the environment, warrants further study on setting of the Still-face procedure. The use of the Still-

face procedure allowed observation of mother-infant interaction in a relatively standardized way over a short period of time. A limitation in the use of the Still-face procedure, however, is that it does not reflect a natural interaction between infant and parent. Mothers may experience the Still-face procedure as somewhat stressful and discomforting for their child (Mayes, Carter, Egger, & Pajer, 1991). It may change their behavior during the reunion episode, when they are reacting to the stress and changes in behavior of the infant as well as to their own reaction to the Still-face procedure. Mothers in the intervention group, however, may have become better able to deal with a special situation like this, because they received information on how to respond to the behavioral cues of their infant by adjusting the environment and offering co-regulatory support.
The findings in this study were limited. Nonetheless we think they provide important information on maternal sensitivity and exploratory behavior of the infants and the effect of the IBAIP. Although IBAIP may not have led to robust changes in mother-infant interaction, the information on our subtle findings is important in view of the developmental improvement of the intervention infants. Indeed, given the huge number of moment by moment interchanges between infants and their mothers even small and subtle changes can accumulate and lead to large developmental differences. Further study on the effect of IBAIP on mother-infant interaction should be done to evaluate what part of the intervention procedure may affect improvements in maternal sensitivity that may contribute development enhancing effect of IBAIP for preterm infants.

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3

Maternal psychological distress in the first two years after very preterm birth and early intervention

'Nadat T. thuis was gekomen en het met hem goed ging, stortte ik in. Natuurlijk had ik toen om hulp kunnen vragen, maar ik had het gevoel dat we iets in te halen hadden samen. Ruim een half jaar ben ik doorgegaan, was eigenlijk nog ziek. Ik ben mezelf een tijd helemaal kwijt geweest. Ik kreeg hyperventilatie en zat zelfs thuis van mijn werk. Niemand begreep het. Ik hoefde niet constant troostende armen om me heen, maar een beetje begrip was wel fijn geweest.'

MATERNAL PSYCHOLOGICAL DISTRESS IN THE FIRST TWO YEARS AFTER VERY PRETERM BIRTH AND EARLY INTERVENTION

Meijssen, D.E., Wolf, M.J.M.A.G., Koldewijn, K., Van Baar, A.L. & Kok. J.H.

Abstract

Preterm delivery may have a strong impact on mothers. In a multicenter randomized controlled trial, including very preterm infants (<32 weeks and/or <1500 grams), the effect of the Infant Behavioral Assessment and Intervention Program (IBAIP) on maternal psychological distress at 6, 12 and 24 (corrected) months after preterm birth was assessed. The program is designed to assist parents to support and enhance their infant's regulatory competence and development.

86 infants and their parents were randomly assigned to the intervention group and 90 to the control group. Maternal psychological distress was assessed with the General Health Questionnaire.

In general, the mothers reported high levels of psychological distress, especially during the first 6 months after discharge from hospital when 56% had clinical scores. No differences were found in maternal psychological distress between the intervention and control group. Early intervention to decrease maternal distress in mothers of preterm infants is warranted.

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INTRODUCTION

Although most very preterm infants nowadays survive, many experience developmental problems. About 11% of the very preterm (< 32 weeks) or very low birth weight infants (< 1500 gram) in the Netherlands develops cerebral palsy and about 57% experiences some difficulty in motor, sensory, cognitive or behavioral development at five years of age (de Kleine et al., 2007). Next to medical complications also psychosocial risk factors like maternal psychological distress and sociodemographic characteristics influence developmental outcome. The development of preterm infants is found to be positively associated with maternal education (Singer et al., 1999) and negatively with the mothers' amount of daily stress (Murray et al., 1999).

After hospital discharge caregiving of the fragile preterm infant very often places a burden on the parents, particularly on the mothers who in general play a major role in caregiving (Eisengart, Singer, Fulton, & Baley, 2003). Moreover, the infant's premature behavior with atypical responses to parental interaction may further complicate parenting (Goldberg & DiVitto, 1995; Schmücker et al., 2005). Although much is known about consequences of prematurity on infant development, little is known about the consequences on maternal psychological distress. Results from the small number of studies are conflicting. Within the first week after delivery no differences were found in maternal depression between mothers of preterm and term born babies (Madu & Roos, 2006). However, in the first year after preterm birth higher levels of psychological distress, traumatic and depressive symptoms were indicated by mothers of preterm children compared to mothers of term infants (Kersting et al., 2004; Singer et al., 1999; Singer et al., 2003). Kersting et al. (2004) reported more traumatic and depressive symptoms from the first week on until 14 months after birth in mothers of very preterm infants compared to mothers of term born infants. Moreover, between 26% and 41% of the mothers reported posttraumatic stress symptoms at 18 months after a premature delivery (Pierrehumbert, Nicole, Muller-Nix, Forcada-Guex, & Ansermet, 2003).

Preventive intervention programs after discharge from the hospital, focusing on the parent-infant relationship and infant development, have shown to be effective in enhancing preterm born infants' cognitive development short term (Spittle, Orton, Doyle, & Boyd, 2007). A review on early interventions for high-risk infants focussing on improvements in parent adaptation and parenting behavior, showed, despite heterogeneity in outcomes and content, a consistent pattern of positive effects on maternal adaptation (e.g., lower anxiety and less depressive symptoms) and parent-

child interaction (Deater-Deckard & Bulkley, 2000). Studies on a modified version of the Mother-Infant Transaction Program (MITP) showed reduced parenting stress at 6, 12 and 24 months, but the MITP failed to enhance infant development (Kaaresen, Ronning, Ulvund, & Dahl, 2006; Kaaresen et al., 2008). The quality of the infants' functioning and development may also be important for maternal psychological distress.

The Infant Behavioral Assessment and Intervention Program (Hedlund, 1998) assists parents to support and enhance their infant's self-regulatory competence, to engage in social and environmental interactions without distress, to reinforce their infants' motivation and autonomy to explore, and to learn from social and external information. We have shown in a randomized multicenter controlled trial that the IBAIP enhances the mental, motor and behavioral outcome of very preterm infants(<32 weeks and/or <1500 grams) at six months corrected age (Koldewijn et al., 2009) In addition we found an increase in the number of sensitive maternal interaction behaviors with the infant (Meijssen et al., in press). Although the intervention program was primarily aimed at improvement of the infants' behavior and development, a secondary effect of this program might be a reduction in maternal distress. To study this secondary outcome of the intervention program and to contribute to the existing contradictory literature on the impact of preterm delivery on psychological distress, we examined maternal psychological distress during the first two years after the initial intervention.

METHOD

Participants

A randomized controlled trial was carried out in Amsterdam, the Netherlands. Two hospitals with level III NICUs and five city hospitals participated in this trial. All infants with a gestational age of <32 weeks and/or a birth weight of <1500 grams, admitted to one of these seven hospitals and whose parents were living in Amsterdam, were eligible for the study if they survived to a post menstrual age of 32-34 weeks. Infants with severe congenital abnormalities, infants whose mothers had a documented history of illicit drug use or severe physical or mental illness, infants from non-Dutch speaking families for whom we could not arrange an interpreter, and infants who participated in other trials on post discharge management were excluded from the study. At 35-38 weeks post menstrual age, the participating infants were randomly assigned to the intervention group or the control group. The randomization was computer-generated at the Academic Medical Center and stratified

for gestational age (<30 and ≥30 weeks) and recruitment site; twins were always assigned to the same group.
Recruitment took place from January 2004 to April 2006; 315 infants were eligible for the study. The parents of 38 infants refused participation, 11 infants died before recruitment, 38 infants were excluded and 52 participated in another post-discharge trial. Finally, 176 infants were left (151 families) for randomization; 86 infants and their 72 parents were assigned to the intervention group and 90 infants and their 79 parents to the control group (for a detailed description see, Koldewijn et al., 2009).
In addition to the preterm control group, data were available from a reference group of 40 infants born term and their mothers and surveyed by the GHQ questionnaire at 6 months of age. They were selected through well baby clinics in 2006 in order to evaluate maternal distress in mothers of young term born infants with the GHQ.
The Medical Ethics Committee of all hospitals involved in this trial approved the study. This trial is registered with controlled-trials.com, number ISRCTN65502576. All participating parents signed informed consent.

The intervention

The IBAIP is a post-discharge preventive intervention program for infants up to the developmental age of 6-8 months and their parents. It is based upon the conceptual framework of Heidelise Als that underlies the Newborn Individual Developmental Care and Assessment Program (NIDCAP) (Als, 1986). The goal of the IBAIP is to enhance the infant's social and environmental interactions without distress, reinforcing the infant's motivation and autonomy to explore and to learn from interactions. It is based on the assumption that the parent's availability and adequate responsiveness strengthens the infant's regulatory competence and development. The intervention is guided by the Infant Behavioral Assessment (IBA) (Hedlund & Tatarka, 1988). The IBA is an observational tool which helps the interventionist to make parents aware of their baby's responses to information.
The infants in the intervention group received 6-8 home visits (of approximately one hour) by the interventionists, who were all experienced IBAIP trained pediatric physical therapists. Infants in both groups received standard care, which consisted of regular visits to the pediatrician in the local outpatient pediatric clinic.

Measures

Psychological distress was assessed with the 28-item version of the General Health Questionnaire (Goldberg & Williams, 1988). The GHQ-28 results in

a total score and four subscale scores: health perception (somatisation); anxiety and insomnia; social dysfunction; and depression. Each item has four answering categories. The Likert's score (item score 0, 1, 2, 3) was used for calculating the subscale scores, and the GHQ score (item score 0, 0, 1, 1) was used for calculating the total score. This total score can be used to estimate the prevalence of clinically important psychological distress with a cut-off point of ≥ 5. In the Dutch norm population (consisting of female adults between 18 and 44 years of age) 39% scored 5 or above (Koeter & Ormel, 1991).
The psychometrical properties of the GHQ-28 as used in the Dutch population are reported to be highly satisfactory with a Cronbach's alpha of 0.94 (Koeter & Ormel, 1991).

Procedure
The preterm infants and their parent(s) were invited to come to the outpatient clinic of our hospital or were visited at home for a physical and developmental examination at the infants' corrected age of 6, 12 and 24 months. Before these visits the GHQ-28 was sent to the parents and they were asked to complete the questionnaire and to hand it over to the assessor. The term infants and their parent(s) also received the GHQ-28 by mail before a home visit. The parent spending the most time with the infant was asked to fill in the questionnaire.

Statistical analyses
Characteristics of the intervention and control groups were compared with t-tests and Chi square tests. A small number of father respondents (five at 6 months, four at 12 and five at 24 months), was excluded from the analyses to increase homogeneity of the sample. Correlational analyses were used to identify associations between GHQ clinical outcomes and medical risk factors. Analyses of variance for repeated measures were performed on the GHQ total score to analyze differences in maternal psychological distress between the intervention and control group over time (at 6, 12 and 24 months corrected age of the infants). Analysis of covariance adjusted for differences in socio-demographic variables (maternal education and country of birth) was performed to compare the GHQ scores of the mothers in the preterm group at 6 months to those of the reference group mothers. One-sample t-test was used to compare the results of the reference group to the norm scores of the Dutch female population. Further explorations were done within the preterm group in relation to family circumstances that have been reported to influence maternal psychological distress and infant development (Miles, Holditch-

Davis, Schwartz, & Scher, 2007; Singer et al., 1999; Davis, Edwards, Mohay, & Wollin, 2003). These circumstances regarded single mothers versus mothers living with a partner, low versus highly educated mothers, mothers born in the Netherlands versus mothers born abroad. All statistical analyses were performed using version 15.0.1 of the Statistical Package for Social Sciences (SPSS 15.0.1, Chicago, IL, USA). An alpha level of 0.05 was used for all tests of significance.

RESULTS

Table 1a and 1b show the sociodemographic characteristics of the 125 mothers of very preterm infants (64 interventions vs. 61 controls) and 40 mothers of term infants who answered the questionnaire at 6 months and the neonatal characteristics of their 147 preterm infants (77 interventions vs. 70 controls) and 40 term infants. Maternal response rates were 86%, 76% and 62% at 6, 12 and 24 months, respectively. Differences were found between respondents and non respondents at 6 months on the following characteristics: Dutch speaking, educational level and born in the Netherlands. In the preterm control group more Dutch speaking (X^2=9.43, p=0.00) and high educated mothers (X^2=6.27, p=0.01) answered the GHQ-28 and in both the intervention and control group most of the respondents were born in the Netherlands (X^2=21.38, p=0.00).
No significant differences were found between the intervention and the preterm control group with respect to neonatal characteristics (gestation, birth weight, infant gender, multiple birth, artificial ventilation, intraventricular hemorrhage and periventricular leukomalacia) and maternal background (age, parity, mother born in the Netherlands, mother speaking Dutch, marital status, maternal education and work status). However, differences were found between the preterm groups and the term group; the term group mothers were higher educated (X^2=12.87, p=0.00) and they were more often born in the Netherlands (X^2=15.77, p=0.00). Therefore, the analyses comparing the results of the mothers of preterm and term children were corrected for these differences.

Within the preterm groups correlational analyses showed no association between gestation and maternal psychological distress at 6 (r=-0.01, p=0.89) and 24 (r=0.13, p=0.23) months after term date. However, a weak linear relationship was found at 12 months (r=0.23, p=0.02). Therefore, gestation was used as a covariate in the analyses of variance for repeated measures. Correlation coefficients between birth weight and GHQ-28 scores at 6, 12 and 24 months after term date were not significant (r=0.14,

Table 1a Sociodemographic characteristics of intervention and control group mothers

	Intervention (n=64)	Control (n=61)	Term (n=40)
Sociodemographic characteristics			
Maternal age (year), mean (SD)	32.1 (5.4)	32.7 (5.0)	32.3 (3.6)
Firstborn child	45 (73%)	36 (63%)	17 (42%)
Mother born in Netherlands	37 (58%)	37 (62%)	38 (95%)
Mother speaking Dutch	56 (88%)	55 (92%)	40 (100%)
Marital status			
Single mother	8 (13%)	5 (9%)	0
Living with a partner	53 (84%)	52 (90%)	40 (100%)
Maternal education			
No high school graduate	22 (34%)	21 (35%)	2 (5%)
High school graduate	42 (66%)	39 (65%)	38 (95%)
Working mother	44 (70%)	40 (69%)	32 (80%)
Infant day care	10 (16%)	16 (28%)	20 (50%)

Table 1b Neonatal characteristics of preterm and term infants

	Intervention (n=77)	Control (n=70)	Term (n=40)
Neonatal characteristics			
Gestation (wk), mean (SD)	29.4 (2.1)	29.9 (2.1)	39.9 (1.7)
Birth Weight (g), mean (SD)	1236 (338)	1304 (340)	3486 (587)
Gender infant: male/female, n	44/33	31/39	17/23
Twins/triplets, n (%)	27 (35%)	24 (34%)	0
Artificial ventilation n (%)	38 (49%)	24 (34%)	-
Intraventricular hemorrhage (IVH) n (%)	18 (25%)	9 (13%)	-
IVH grade I +II / III + IV	14 / 4	7 / 2	-
Periventricular leukomalacia (PVL) n (%)	10 (13%)	9 (13%)	-
PVL grade 1 / 2+3	9 / 1	7 / 2	-

Numbers are given as numbers of infants unless stated otherwise

Table 2 Mean GHQ scores and standard deviations at 6, 12 and 24 months corrected age

	6 months		12 months		24 months		Group[1]	Time[2]	Group x Time[3]
	Intervention (n=64) M (SD)	Control (n=61) M (SD)	Intervention (n=57) M (SD)	Control (n=47) M (SD)	Intervention (n=49) M (SD)	Control (n=39) M (SD)	F	F	F
GHQ-28 total score	7.36 (5.71)	6.30 (5.88)	5.20 (5.81)	5.29 (6.58)	3.80 (4.88)	4.31 (5.88)	0.36	3.73*	0.54
Somatisation	7.55 (4.09)	6.85 (4.47)	6.15 (4.18)	6.24 (4.30)	5.35 (3.31)	5.44 (4.30)	1.42	2.98	0.36
Anxiety and insomnia	7.09 (4.64)	6.64 (5.37)	5.79 (4.86)	5.90 (5.20)	5.20 (4.46)	5.43 (5.39)	0.24	4.20*	0.23
Social dysfunction	8.39 (3.50)	7.23 (3.22)	7.31 (2.76)	7.65 (2.45)	7.06 (2.80)	6.97 (2.43)	1.33	2.95	0.90
Depression	1.86 (2.72)	1.66 (2.62)	1.51 (2.75)	1.73 (3.19)	0.67 (1.72)	0.79 (2.10)	2.91	3.60*	0.91

[1] Group: tests differences between intervention and control group
[2] Time: tests differences over time
[3] Group x Time: tests interaction effect in change over time of both groups
Analyses of variance for repeated measures (corrected for gestation)
**p≤.05*

r=0.01 and r=0.08, respectively). The mothers of the infants who needed artificial ventilation did not differ in GHQ-28 scores over time (F (3, 96)=1.76, p=0.16; Wilks' Lambda=0.95), neither did the mothers of the infants with an intraventricular hemorrhage (F(3, 95)=0.35, p=0.79; Wilks' Lambda=0.99) or periventricular hemorrhage (F(3, 95)=0.48, p=0.70; Wilks' Lambda=0.98).

Table 2 presents the mean GHQ-28 scores and standard deviations in the intervention and control group mothers at 6, 12, and 24 months. No differences were found in maternal psychological distress between both groups at 6, 12 and 24 months as shown by analyses of variance for repeated measurements. Separate analyses of variance for repeated measures in the intervention group showed a significant decrease in GHQ-28 total scores between 6 and 24 months (mean difference=2.78, SE=.86; p=.01). In the control group no significant decrease was found (mean difference=1.72, SE=.90; p=.20).
The percentage of women who scored above the clinical cut-off level of ≥ 5 did not differ between the intervention and control preterm group at 6 (X^2=2.25, p=0.13), 12 (X^2=0.20, p=0.66) and 24 months (X^2=0.01, p=0.91).

The GHQ scores of the total group of mothers of preterm children were very high at 6 months, which could also be related to the special circumstances in providing care for a young child. Hence we performed a further exploration and compared the results of the mothers of the preterm infants to those of mothers of term born infants at 6 months (Table 3). Analysis of covariance adjusted for maternal education and country of birth showed a significant difference on the GHQ total score (F(1,182)=4.01, p=0.05); mothers of preterm children reported more psychological distress

Table 3 GHQ scores at 6 months' corrected age for preterm and term group mother

	Preterm n=147 M (SD)	Term n=40 M (SD)	F
GHQ-28 total score	6.74 (5.82)	4.05 (3.52)	4.01*
Somatisation	7.11 (4.39)	5.22 (2.73)	2.63
Anxiety and insomnia	6.86 (5.03)	4.90 (3.68)	3.25†
Social dysfunction	7.67 (3.43)	6.92 (2.03)	0.86
Depression	1.71 (2.60)	0.70 (1.62)	2.88†

Analysis of covariance corrected for maternal education and country of birth
**p≤.05, †p≤.10*

compared to the mothers of term born infants, whereas the results of the mothers of the term born infants did not differ from the population norms for women as analysed using a T-test for one sample (T(39)=-1.15, p=0.26).

Subgroup analyses

In order to explore results of specific subgroups, the GHQ scores were also analyzed in relation to family status (living with partner or single mother), educational level and country of birth. No differences in maternal psychological distress were found among the subgroup mothers in the intervention and control group. Separate analyses were also done for the mothers with clinical scores on the depression subscale (GHQ-D ≥ 5): 17 mothers (14%) reported symptoms of serious depression, however no differences were found between the 10 intervention and 7 control group mothers at 6 months (X^2=0.46, p=0.50) and (7 vs. 9) at 12 months (X^2=0.00, p=0.94). At 24 months only 2 mothers (2%) in the preterm group reported serious depression symptoms. In the term group only 1 mother (2.5%) reported symptoms of serious depression at 6 months. These percentages are not significantly different.

Combining the outcome of the total group of mothers with preterm infants (both intervention and control group), 56% of the mothers reported clinical levels of psychological distress at 6 months in comparison to 35% of the term group mothers (X^2=5.80, p=0.02), which was comparable to the 39% of the general Dutch female population (Koeter et al., 1991). The mean GHQ-28 total score in the combined group of preterm infants at 6 months was 6.74 (SD=5.82, range 0 – 26), significantly higher (T(146)=4.27; p=0.00) than the mean GHQ total score of 4.69 in the general Dutch female population (Koeter et al., 1991) and the mean GHQ total score of 4.05 in the term group. At 12 months the mean GHQ-28 total score of all the mothers was 5.24 (SD=6.14) compared to 4.69 in the general Dutch female population (T(115)=0.66; p=0.51) and 38% of the mothers had GHQ-28 scores in the clinical range. At 24 months the mean GHQ-28 total score of all the mothers was 4.02 (SD=5.32) and 35% of the mothers had GHQ-28 scores in the clinical range, which is quite comparable to the general Dutch female population.

DISCUSSION

Using the General Health Questionnaire at 6, 12 and 24 months after term date, no differences in maternal psychological distress were found between mothers of infants who received IBAIP after discharge from the Neonatal Intensive Care Unit, compared to mothers whose infants did not

receive this intervention. In general, all mothers of preterm infants reported much psychological stress, with more than half (56%) of the mothers scoring above the clinical cut-off level at 6 months after term date. In the group of mothers of term babies only 35% scored above the clinical cut-off level, which clearly demonstrates the impact of delivering a very preterm born baby. Especially anxiety and insomnia and depression were reported by the mothers of preterm children 6 months after term date. Two years after birth most mothers seem to have found a way to cope with most of their psychological distress complaints, because the mean GHQ-28 score dropped below the clinical level of 5 at 24 months; 35% of the mothers then reported clinically relevant psychological distress, which is comparable to mothers of term babies and the general Dutch female population. This indicates maternal resilience while caring for a vulnerable and fragile preterm infant. The ability of these mothers to adapt and to overcome their psychological imbalance two years after delivering a very preterm born baby is admirable.

It may be important to realize that a number of the mothers in our study may already have been psychologically distressed before childbirth, as previous studies have found an association between psychological stress and preterm delivery (Field, Diego, & Hernandez-Reif, 2006; Mulder et al., 2002) but intra-individual stability in psychological distress during pregnancy and two years after delivery has also been found (Dipietro, Costigan, & Sipsma, 2008). Mothers in our preterm sample may already have had a higher baseline of distress and depressive complaints compared to the general population, which could have contributed to the high GHQ-28 scores at 6 months. However, this level of distress normalizes after the first year.

Although it is reassuring that the mothers are able to recover in the course over time, it is important to realize that mother-infant attachment processes during the first 6 months take place under conditions of high maternal stress, which is a cause of concern for the infant's early socio-emotional development or attachment quality during that early period. It is therefore important to pay attention to maternal psychological distress after very preterm birth specifically in the first six months after discharge. Especially the 14% of mothers with depression symptoms in the clinical range demand specific attention and early appropriate support. In mothers of term babies in this study and in the literature only 0-2% of the mothers reported similar amounts of depression at 6 months (Skari et al., 2002). The use of standardized questionnaires as part of the parental counseling process may improve case identification, leading to early intervention that ideally should start in the NICU and continue after discharge.

Comprehensive early intervention, supporting infant development, mother-infant interaction and maternal psychological well being, is needed. The IBAIP did effectively support infant development (Koldewijn et al., 2009) and sensitive mother-infant interaction (Meijssen et al., in press). The program was developed to provide parents support in order to help them achieve the best potential for their children (Hedlund, 1998). The results from this study suggest that additional maternal psychological support is warranted. The outcome of a questionnaire like the GHQ could be used explicitly during the intervention sessions, or extra sessions focused upon the individual mothers' concerns could be added to IBAIP. Pre-delivery counseling policies that have been implemented in some hospitals for mothers who are at risk of delivering preterm infants, may be helpful (Allen, 2002). By preparing parents for upcoming events and potential complications, they may be less emotionally distressed after their preterm baby is born and better able to cope with the situation. Specific psychological therapy may be needed in cases of an actual post traumatic stress disorder (Bisson & Andrew, 2007). However, no consensus exists on the best way to support parental psychological well being and to prevent psychological distress after preterm birth (Deater-Deckard et al., 2000). From our study it appears that the incorporation of methods to disclose adverse feelings of the mothers like anxiety, insomnia and depression, and the inclusion of appropriate supports, including extra support to the mothers, could prove to be important extensions of the original IBAIP.
A limitation to this study is the disproportionate attrition from our original sample of families of lower educated mothers and mothers born outside the Netherlands, but it has also been seen in other long-term follow-up studies (Wolke, Ratschinski, Ohrt, & Riegel, 1994). However, the possibility that this has contributed to a positive bias of our results seems small, as such background variables were not related to psychological distress in the responding parents. Furthermore, the number of respondents to the questionnaires at 12 and 24 months turned out to be lower in the control group than in the intervention group. This may indicate an intervention effect in itself, as intervention mothers might have felt more dedicated to the study and therefore cooperated better. The total drop out rate of 38% at 24 months definitely reduces the power of the results.

In conclusion, mothers of very preterm infants report high levels of psychological stress especially in de first half year after discharge from hospital despite IBAIP intervention, but these high stress levels decreased as the children grow older.

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4

Parenting stress in mothers after very preterm birth and the effect of the Infant Behavioral Assessment and Intervention Program

'Al dat testen en elke keer weer horen dat het niet goed gaat, dat hij achterloopt, terwijl ik zo trots was dat hij zichzelf had omgedraaid in de box. Hij is specialer en knapper dan andere kinderen. Hij heeft het echt allemaal doorstaan en overleefd. Ik heb daar echt bewondering voor. Respect is niet het goede woord. In het ziekenhuis zeiden ze altijd het is een vechtertje, en daar moest ik een beetje om lachen, maar het is wel waar.'

PARENTING STRESS IN MOTHERS AFTER VERY PRETERM BIRTH AND THE EFFECT OF THE INFANT BEHAVIORAL ASSESSMENT AND INTERVENTION PROGRAM

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Abstract

Objective: Purpose of this study was to examine maternal parenting stress as a secondary outcome of the Infant Behavioral Assessment and Intervention Program (IBAIP).

Method: In a randomized controlled trial 86 very preterm infants and their parents were assigned to the intervention group and 90 to the control group. Maternal parenting stress was assessed with the Dutch version of the Parenting Stress Index at 12 and 24 months post term.

Results: The intervention group mothers reported less parenting stress, as they perceived their infants as happier and less distractible. However, they also reported more feelings of social isolation compared to the control mothers.

Conclusions: The IBAIP appears to have made mothers more satisfied about their infants' mood and distractibility, but also may have evoked more feelings of social isolation.

Submitted for publication

INTRODUCTION

Parenting stress is caused by a mismatch between the perceived demands of parenting and the resources available to meet those demands (Abidin, 1995). Increased parenting stress is a risk factor for maladaptive parenting (Kazdin & Whitley, 2003), as well as higher levels of child disruptive behavior problems (Barry, Dunlap, Cotten, Lochman, & Wells, 2005). Parent as well as child characteristics influence parenting stress in complex ways. Risk factors are single parenthood (Ricciuti, 2004; Williford, Calkins, & Keane, 2007), parental psychopathology (Misri, Reebye, Milis, & Shah, 2006; Sheinkopf et al., 2006; Williford et al., 2007), difficult temperament (Ostberg & Hagekull, 2000) and regulation difficulties of the child (Calkins & House, 2004).

The demands of parenting a young child could be especially stressful for parents if the child is born very preterm, which results in an increased risk for morbidity and potential developmental delay (de Kleine et al., 2007). Reports on the impact of preterm birth on parenting stress are inconsistent. Some studies report more parenting stress in mothers of high-risk very low birth weight infants (Auslander, Dvorah, & Arad, 2003; Drotar et al., 2006; Singer et al., 1999; Singer et al., 2007; Taylor, Klein, Minich, & Hack, 2001). Other studies find similar levels of parenting stress in parents of term or preterm infants (Candelaria, O'Connel, & Teti, 2006; Saigal, Burrows, Stoskopf, Rosenbaum, & Streiner, 2000). Intervention programs focused upon improving preterm infants' development, could also affect parenting stress, either as a direct result from extra attention for the family and child, or through improvements in development of the children. A review on early intervention in high-risk infants focussing on parent adaptation and parenting behavior showed, despite heterogeneity in outcomes and content, a consistent pattern of positive effects on maternal adaptation (e.g., less anxiety and depressive symptoms) and parent-child interaction (Deater-Deckard & Bulkley, 2000). Studies on a modified version of the Mother-Infant Transaction Program (MITP) (Rauh, Nurcombe, Achenbach, & Howell, 1990) showed reduced parenting stress at 6, 12 and 24 months in a group of very preterm infants (Kaaresen, Ronning, Ulvund, & Dahl, 2006; Kaaresen et al., 2008). Unfortunately the MITP did not enhance infant development until that age.

We studied an intervention program that focuses on improvement of the self-regulatory competence of very preterm infants. Self regulatory competence is thought to be important for the infant's social interactive and exploratory capacities and opportunities, which are necessary for learning processes (Bronson, 2000). In a randomized controlled trial carried

out in Amsterdam, the Netherlands, was shown that the Infant Behavioral Assessment and Intervention Program (IBAIP)(Hedlund, 1998) enhances the mental, motor and neurobehavioral outcome of very preterm infants at 6 months corrected age (Koldewijn et al., 2009). Also mother-infant interaction and maternal sensitivity was found to be improved in the intervention group at this age (Meijssen, Wolf, Koldewijn, Houtzager, Van Wassenaer, Tronick, Kok & Van Baar, in press). However, no effect was found on maternal psychological distress as reported on the General Health Questionnaire (Meijssen, Wolf, Koldewijn, Van Baar & Kok, in press). As a secondary outcome of the IBAIP, parenting stress may also have been affected. We hypothesized that the strength based approach of the IBAIP and the emphasis on parental empowerment would be associated with less parenting stress.

METHOD

Participants

A randomized controlled trial was carried out in Amsterdam, the Netherlands, using the IBAIP to support and enhance infant's regulatory competence, maternal sensitivity and mother-infant interaction in very preterm infants. Two hospitals with level III NICUs and five city hospitals participated in this trial.

All infants with a gestational age of <32 weeks and/or a birth weight of <1500 grams, admitted to one of these seven hospitals and whose parents were living in Amsterdam, were eligible for the study if they survived to a post menstrual age of 32-34 weeks. Infants with severe congenital abnormalities, infants whose mothers had a documented history of illicit drug use or severe physical or mental illness, infants from non-Dutch speaking families for whom we could not arrange an interpreter, and infants who participated in other trials on post discharge management were excluded from the study. At 35-38 weeks post menstrual age, the participating infants were computer-generated randomly assigned and stratified for gestational age (<30 and ≥30 weeks) and recruitment site to the intervention group or the control group. Twins were always assigned to the same group.

Recruitment took place from January 2004 to April 2006; 315 infants were eligible for the study. The parents of 38 infants refused participation, 11 infants died before recruitment, 38 infants were excluded and 52 participated in another post-discharge trial. Finally, 176 infants were left (151 families) for randomization; 86 infants and their 72 parents were assigned to the intervention group and 90 infants and their 79 parents to

the control group (for a detailed description see, Koldewijn et al., 2009). The Medical Ethics Committee of all hospitals involved in this trial approved the study. This trial is registered with controlled-trials.com, number ISRCTN65502576. All participating parents signed informed consent.

Intervention

The IBAIP is a post-discharge preventive intervention program for infants up to the developmental age of 6-8 months and their parents. It is based upon the conceptual framework of Heidelise Als that underlies the Newborn Individual Developmental Care and Assessment Program (NIDCAP) (Als, 1986). The goal of the IBAIP is to enhance the infant's social and environmental interactions without distress, reinforcing the infant's motivation and autonomy to explore and to learn from interactions. It is based on the assumption that the parent's availability and adequate responsiveness strengthens the infant's regulatory competence and development. The intervention is guided by the Infant Behavioral Assessment (IBA) (Hedlund & Tatarka, 1988). The IBA is an observational tool which helps the interventionist to make parents aware of their baby's responses to information.

The infants in the intervention group received 6-8 home visits (approximately one hour) by the interventionists, who were all experienced IBAIP trained pediatric physical therapists. Infants in both groups received standard care, which consisted of regular visits to the pediatrician in the local outpatient pediatric clinic.

Instruments

Parenting stress was assessed with the Dutch version of the Parenting Stress Index (Abidin, 1983): the Nijmeegse Ouderlijke Stress Index (NOSI) (de Brock, Vermulst, Gerris, & Abidin, 1992). The NOSI (applied at 24 months corrected age) measures parenting stress on the basis of 123 items in several subscales related to the parent: sense of competence, restriction of role, attachment, depression, parent's health, social isolation, and relationship with spouse; and several subscales related to the child: adaptability, mood, distractibility/hyperactivity, demandingness, 'reinforces parent' and acceptability. Higher scores indicate more stress. The NOSI can be used in children between 2 and 13 years of age.

The internal consistency of all NOSI subscales meet the criterion of an alpha coefficient ≥0.70; the parent and child domain scales as well as the total scale have an alpha coefficient >0.90 which indicates good reliability. The criterion validity and the concurrent validity of the test are qualified as satisfactory; indications for discriminating validity have been

found (de Brock et al., 1992).
At 12 months the short version of the Nijmeegse Ouderlijke Stress Index (NOSIK) (de Brock et al., 1992) was used which consists of 25 parenting stress related statements (the items that performed best on factor analysis in the NOSI complete version) with answers on a 5 point Likert scale ranging from 1 (totally disagree) to 5 (totally agree) resulting in a total stress score. The NOSIK can be used in children aged 1 and older. The parent spending the most time with the infant was asked to fill in the questionnaires.
Psychological distress of the mothers was assessed with the 28-item version of the General Health Questionnaire (Goldberg & Williams, 1988). The GHQ-28 measures total psychological distress (health perception, anxiety and insomnia, social dysfunction, and depression). The psychometrical properties of the GHQ-28 as used in the Dutch population are reported to be highly satisfactory with a Cronbach's alpha of 0.94 (Koeter & Ormel, 1991).

Statistical analyses
Characteristics of the intervention and control group were compared with analysis of variance or Chi square tests. A small number of father respondents (7 at 12 and 8 at 24 months), was excluded from the analyses to increase homogeneity of the sample.
Correlational analyses and analyses of variance were used to identify associations between the NOSI(K) and neonatal and sociodemographic characteristics. Factors associated with the outcome (NOSIK or NOSI) and factors differing significantly between the intervention and control group, were used as covariates in the analyses of variance. Analysis of covariance was performed for group comparison on the total score of the NOSIK. Multivariate analyses of covariance were performed for group comparisons on the NOSI subscales. The number of pairwise comparisons was controlled for by Bonferroni post hoc tests.
All statistical analyses were performed using version 15.0.1 of the Statistical Package for Social Sciences (SPSS 15.0.1, Chicago, IL, USA). An alpha level of 0.05 was used for all tests of significance.

RESULTS

Respondents
Between 6 and 24 months corrected age, parents of 1 intervention infant withdrew and 2 intervention infants moved abroad; in the control group 2 infants died, 2 infants moved abroad, 3 infants were withdrawn, and 5 infants were lost to follow up; thus leaving 83 (97%) intervention infants

and 79 (90%) of the surviving control infants available for follow-up at 12 months and respectively 83 and 78 at 24 months.
123 mothers completed the NOSIK at 12 months (response rate 79%) and 103 mothers completed the NOSI at 24 months (response rate 67%). At both times the response rate was less in the control group. The NOSIK was answered by 86% of the intervention group mothers versus 63% of the control group (X^2=11.92, p=0.00). The NOSI was answered by 74% of the intervention group versus 56% of the control group (X^2=6.29, p=0.01). More Dutch speaking mothers, mothers who were born in the Netherlands and high educated mothers responded to the NOSIK as well as the NOSI in both the intervention and control group. The non responders in the intervention and control group did not differ, except for two of the neonatal characteristics of their children. At 12 months more infants of the non responders in the control group had had intracerebral hemorrhage (9 (69%) versus 5 (24%); X^2=7.73, p=0.00) in the intervention group. At 24 months the infants of the non responders in the control group had a lower birth weight (mean difference=186.10, se=67.52, T(84)=-2.76, p=0.01).

The sociodemographic characteristics of the 103 mothers (60 intervention vs. 43 control group) that answered the questionnaire at 24 months and the neonatal characteristics of their infants are shown in table 1a and 1b. Despite the randomized control design and similar to the complete study group (Koldewijn et al., 2009), important differences in neonatal characteristics to the disadvantage of the intervention group infants were found: intervention group infants were more often born before 28 weeks, had a lower mean birth weight, were more often oxygen dependent at 36 weeks gestational age and had more septic episodes than the infants in the control group.
At 12 months, neonatal characteristics of the infants of the mothers that responded to the NOSIK also differed between the intervention and control group. A lower mean gestational age, more children that needed oxygen at 36 weeks gestational age, more septic episodes and occurrence of intracerebral hemorrhage, as well as more multiplets were found in the intervention versus the control group.
No differences were found between intervention and control group mothers in sociodemographic characteristics at both ages. The mothers also did not differ in psychological well being as measured with the General Health Questionnaire (Meijssen et al., in press).

Table 1a Sociodemographic characteristics of intervention and control group mothers

	Intervention (n=60)	Control (n=43)	P
Socio-demographic characteristics			
Maternal age, years, mean (SD)	32.7 (5.4)	32.1 (4.7)	0.62
Firstborn child	40 (66%)	26 (60%)	0.82
Mother born in Netherlands	45 (74%)	33 (77%)	0.48
Mother speaking Dutch	55 (90%)	38 (88%)	0.77
Marital status			
Single mother	7 (11%)	3 (7%)	
Living with a partner	53 (87%)	39 (91%)	0.53
Maternal education:			
No high school graduate	18 (29%)	12 (28%)	
High school graduate	43 (71%)	30 (72%)	0.92

Note: Analyses of variance and Chi-square tests

Table 1b Neonatal characteristics of intervention and control group infants

	Intervention (n=60)	Control (n=43)	P
Neonatal characteristics			
Gestation (wk), mean (SD)	29.5 (2.2)	30.3 (1.9)	0.07
Gestation < 28 weeks	14 (23%)	1 (2%)	0.00
Birth Weight (g), mean (SD)	1238 (339)	1397 (333)	0.02
Gender infant: male/female (%)	57/43	49/51	0.39
Multiplets, n (%)*	21 (34%)	16 (37%)	0.77
Oxygen 36 weeks post menstrual age †	16 (26%)	3 (7%)	0.01
Septic periods	46 (75%)	21(49%)	0.00
Intraventricular haemorrhage (IVH) n (%)▫	15 (25%)	5(12%)	0.09
IVH grade I +II / III + IV	12 / 3	3 / 2	0.45
Periventricular leukomalacia (PVL) n (%)▫	8 (13%)	6 (14%)	0.90
PVL grade 1 / 2+3	7/ 1	4/ 2	0.46

Note: Numbers are given as number of infants unless otherwise stated
Analyses of variance and Chi-square tests
**at least 2 infants survived*
† supplemental oxygen use at 36 weeks post menstrual age is a marker of chronic long disease
▫ Intraventricular haemorrhage (IVH) was defined according to Papile et al. (1983), Periventricular leukomalacia (PVL) according to De Vries, Eken & Dubowitz (1992)

Outcome at 12 months

The mean parenting stress in the intervention group at 12 months was 43.65 (SD=16.84) compared to 41.96 (SD=14.76) in the control group, a non significant difference. A higher gestational age was related to more stress and younger, non-Dutch speaking and single mothers reported more stress. Mothers who reported more psychological distress at 12 months also reported more parenting stress (Table 2).

Analysis of covariance adjusted for neonatal and maternal characteristics associated with the outcome or differing significantly between groups (gestational age, birth weight, oxygen therapy ≥ 36 weeks, septic episodes, intracerebral hemorrhage, family status, multiplets, maternal age, Dutch language speaking and psychological distress (GHQ)) also showed no differences in parenting stress (mean difference=1.16, SE=2.75, p=0.67).

Outcome at 24 months

The mean total parenting stress in the intervention group at 24 months was 225.85 (SD=58.68) compared to 234.98 (SD=53.70) in the control group, a non significant difference. Low educated and single mothers reported more parenting stress. Mothers, who reported more psychological distress when their infants were 12 and 24 months corrected age, also reported more parenting stress at 24 months (Table 2). Group differences on the NOSI subscales were studied with multivariate analysis of covariance adjusted for neonatal and maternal characteristics associated with the outcome or differing significantly between groups (gestational age < 28 weeks, birth weight, oxygen therapy ≥ 36 weeks, septic episodes, family status, maternal education, and psychological distress (GHQ)). This showed an overall difference in parenting stress between the intervention and control group mothers (Willks' λ = 0.68 (F (13, 65)=2.34, p=0.01). In particular the groups differed in infant mood (F(1,77)=4.98, p=0.03), infant distractibility (F(1,77)=5.67, p=0.02), and social isolation (F(1,77)=4.82, p=0.03). Mothers in the intervention group assessed their infants as happier and less hyperactive/distractible compared to the control group mothers. Mothers in the intervention group reported more social isolation (Table 3).

Table 2 Relations between parenting stress and neonatal and sociodemographic characteristics and maternal psychological distress

Total group of mothers (n=103)	NOSIK 12 months	NOSI 24 months
	r	r
Gestation	0.21*	-0.03
Birth Weight	0.04	0.01
Artificial ventilation (days)	-0.10	-0.05
Maternal age, mean (SD), y	-0.37**	-0.11
GHQ-28[1] (12 months)	0.47**	0.25**
GHQ-28 (24 months)	0.09	0.34**
NOSIK		0.34**
	F	F
Gender infant: male/female	0.04	0.02
Multiplets	0.76	1.06
Oxygen 36 weeks post menstrual	0.00	0.51
Intraventricular haemorrhage (IVH)	1.64	0.19
Periventricular leukomalacia (PVL)	2.29	0.20
Firstborn child	0.99	0.64
Mother born in Netherlands	3.50	0.06
Mother speaking Dutch	3.87*	0.36
Marital status	6.34*	5.22*
Maternal education	2.26	3.79*

Pearson correlation coefficient and ANOVA
[1] *GHQ-28: General Health Questionnaire (maternal psychological distress)*
*$p \leq .05$, **$p \leq .01$

Table 3 Mean maternal NOSI scores and standard deviations

	Intervention (n=60) M (SD)	Control (n=43) M (SD)	F	η (effect size)	Adjusted mean difference (95% CI)[a]
Competence	19 (6.67)	22 (7.85)	3.28†	0.03	-1.80 (-4.85 to 1.25)
Restriction of role	16 (6.49)	16 (7.46)	0.08	0.00	0.53 (-2.77 to 3.83)
Attachment	10 (3.89)	10 (3.06)	0.22	0.00	0.00 (-1.70 to 1.70)
Depression	21 (8.56)	22 (8.45)	0.82	0.01	-2.21 (-5.97 to 1.54)
Parent's health	14 (5.10)	14 (5.93)	0.08	0.00	0.14 (-2.13 to 2.41)
Social isolation	13 (5.17)	11 (4.24)	4.30*	0.04	2.35 (0.22 to 4.48)*[b]
Relationship with spouse	15 (6.11)	14 (7.01)	0.55	0.01	2.11 (-0.76 to 4.97)
Adaptability	26 (8.19)	28 (7.61)	1.06	0.01	-1.86 (-5.55 to 1.83)
Mood	14 (5.00)	16 (4.71)	5.15*	0.05	-2.37 (-4.48 to -0.25)*[b]
Distractibility/ hyperactivity	29 (7.53)	32 (8.43)	6.88*	0.07	-4.51 (-8.27 to -0.74)*[b]
Demandingness	17 (7.12)	18 (6.97)	0.09	0.00	-1.20 (-4.25 to 1.85)
Reinforcement	15 (4.55)	16 (4.19)	0.65	0.01	-0.52 (-2.68 to 1.63)
Acceptability	17 (6.37)	17 (4.51)	0.03	0.00	-0.73 (-3.30 to 1.84)

[a] *MANCOVA adjusted for neonatal and demographic variables (gestational age < 28 weeks, birth weight, oxygen therapy ≥ 36 weeks, septic episodes, family status, multiplets, maternal education, and psychological distress (GHQ))*
$p \leq .05$, †$p \leq .10$
[b] *Adjustment for multiple comparisons: Bonferroni*
'Willks' $\lambda = 0.68$ (F (13, 65)=2.34, $p=0.01$; higher scores indicate more parenting stress

DISCUSSION

Mothers who received IBAIP after discharge from the Neonatal Intensive Care Unit, experienced less parenting stress at 24 months corrected age of the children. They experienced their children as happier and less hyperactive/distractible than the control mothers. The strength based and empowering approach of the IBAIP may have supported the mothers in feelings of competence and satisfaction in their children, which might be mirrored in their ratings of the NOSI. The mothers may also have actually observed better concentration and satisfaction in their children. These results are partly in line with the Norwegian study on the MITP intervention (Kaaresen et al., 2008a), that also showed less parenting stress in the intervention group at 24 months. In that study also the mood and distractibility/hyperactivity subscales showed positive intervention effects. A surprising finding was that the intervention mothers felt somewhat more isolated compared to the control group mothers. This may indicate that more responsible and available caretaking may be at the cost of the parent's social life. The IBAIP guided the parents in supporting their infant's self-regulatory competence and aimed to support parents in adjusting the environment to the neurobehavioral needs of the infant. The intervention may have increased the parents' availability and feelings of responsibility towards their child, which may have resulted in less parental social activities. More information from the parents is needed to detect the aspects of the IBAIP which may have contributed to these feelings of social isolation. The IBAIP interventionist should become aware of this possible consequence of the intervention.
Weak to moderate correlations were found between maternal parenting stress and maternal psychological distress at 12 and 24 months. Hence, when assisting parents in coping with the consequences of a preterm birth, it seems important to pay attention to aspects of parenting, as well as to personal feelings or complaints of the parents.

The fact that no intervention effect was found at 12 months, which was earlier in time in relation to the intervention, might be explained by the use of the short version of the NOSI, the NOSIK. It does not measure separate aspects of parenting stress and could miss on the specific aspects that may be important for parents of very preterm infants. Further study concerning parenting experiences in this specific group of parents may benefit from other methods like in depth interviewing to include all aspects of parenting stress.

Unfortunately, the number of respondents to the questionnaires at 24 months turned out to be lower in the control group than in the intervention group. This may indicate an intervention effect in itself, in that the intervention mothers felt more dedicated to the study and therefore cooperated better. The disproportionate attrition from our original sample of families of lower educated mothers and mothers born outside the Netherlands, is unfortunate, but it has also been seen in other long-term follow-up studies (Wolke et al., 1994). The non responders at 12 months had more infants with severe neonatal complications or illnesses. However, the possibility that this has contributed to a positive bias of our results seems small, as such neonatal characteristics were not related to parenting stress in the responding parents.

We conclude that the IBAIP improved some aspects of maternal parenting stress. The mothers assessed their infants as happier and less distractible at 24 months corrected age. However, the mothers in the intervention group also reported more feelings of social isolation, a finding that needs further study.

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5

Maternal attachment representations after very preterm birth and the effect of early intervention

'Ik heb me wel zorgen gemaakt om onze hechting. Ik voelde toch altijd een bepaalde afstand. Niet bewust, maar toch altijd met het idee dat hij nog dood kon gaan of heel erg ziek zou worden... Maar ik merkte dat het goed zat toen ik een keer een mes liet vallen in de keuken en dat ik mijn arm uitstak om het mes op te vangen voordat het op hem zou komen. Dat was wel pas ruim een jaar na zijn geboorte.'

MATERNAL ATTACHMENT REPRESENTATIONS AFTER VERY PRETERM BIRTH AND THE EFFECT OF EARLY INTERVENTION

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Abstract

Background: Very preterm infants are at risk for developmental problems and the mother-infant relationship may be compromised. The purpose of this study was to examine maternal attachment representations 18 (corrected) months after very preterm birth as a secondary outcome of the Infant Behavioral Assessment and Intervention Program (IBAIP). Method: In a randomized controlled trial 86 very preterm infants (<32 weeks and/or <1500 grams) and their parents were assigned to the intervention group and 90 to the control group. Maternal attachment representations were assessed with the Working Model of the Child Interview (WMCI). Results: Seventy-eight mothers were interviewed. In the total group studied, 70% of the mothers had balanced representations, indicating a secure relationship. No differences were found in maternal attachment representations between the intervention and control group. Qualitative content analysis of the answers showed that negative first experiences between the mother and her newborn infant in the hospital and at home are related to nonbalanced attachment representations. Conclusion: The IBAIP in its current form did not affect maternal attachment representations after very preterm birth. Early support for mothers of very preterm born infants to develop a healthy mother-infant relationship is recommended especially for mothers who report negative first experiences.

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INTRODUCTION

A crucial part of infant development is the development of a secure emotional attachment relationship with the primary caregiver (Sroufe, 1988). Premature birth may complicate the development and quality of this attachment relationship. In general the literature is still inconclusive concerning the quality of preterm infants' attachment relationships (Buchheim, Brisch, & Kachele, 1999; Butcher, Kalverboer, Minderaa, van Doormaal, & Wolde ten, 1993; Laganiere, Tessier, & Nadeau, 2003) and possible differences between term and preterm infants. Some studies found more insecurely attached preterm infants compared to term born infants (Mangelsdorf et al., 1996), whereas other studies found comparable numbers (Brisch et al., 2005).

Regarding the outcome of preterm birth, attachment has usually been studied from the infant's perspective. However, after preterm birth, bonding processes of the mothers to their infants may also be at risk (Borghini et al., 2006). Early separation due to the infant's bio-medical complications and treatment results in actual distance between mothers and their preterm newborns (Feldman, Weller, Leckman, Kuint, & Eidelman, 1999). In addition the potential traumatic impact of premature birth may cause stress reactions, like anxiety and depression (Kersting et al., 2004; Kersting et al., 2009; Singer et al., 1999) that may affect maternal feelings and responses towards her infant. Also, the neurobehavioral consequences of neonatal risk factors can affect maturation and functioning of the preterm infant's brain, and influence the infants' responses during mother-infant interaction processes. Several studies have indicated that mother-infant interaction processes differ in dyads with term or very preterm infants (Bozzette, 2007; Eckerman, Hsu, Molitor, Leung, & Goldstein, 1999; Holditch-Davis, Schwartz, Black, & Scher, 2007; Swartz, 2005). Preterm infants show more negative affect and avoid eye-contact compared to term infants (Eckerman et al., 1999), which results in non-optimal responsive parenting (Censullo, 1994).

Maternal bonding processes are reflected in mental representations of attachment. These representations can be understood as mothers' internal subjective experiences of the relationship with their infant (Zeanah & Benoit, 1995). Maternal attachment representations in mothers of very preterm infants may have been affected by preterm delivery and its consequences (Borghini et al., 2006).

Preventive post-hospital discharge interventions directed at interactions between preterm infants and their parents may improve developmental outcome (Spittle, Orton, Doyle, & Boyd, 2007; Bonnier, 2008; Vanderveen, Bassler, Robertson, & Kirpalani, 2009). In a randomized controlled trial

carried out in Amsterdam, the Netherlands, we have shown that the Infant Behavioral Assessment and Intervention Program (IBAIP) (Hedlund, 1998) enhances the mental, motor and neurobehavioral outcome of very preterm infants (<32 weeks and/or <1500 grams)(Koldewijn et al., 2009) and mother-infant interaction and maternal sensitivity at six months of corrected age (Meijssen, Wolf, Koldewijn, Houtzager, Van Wassenaer, Tronick, Kok & Van Baar, in press). As a secondary outcome of the IBAIP, bonding processes may have been affected, as a result of the attention for the mother-infant interaction processes that was addressed during the intervention program. In this study the data are presented of the mothers' attachment representations of their very preterm infants assessed with the Working Model of the Child Interview (Zeanah et al., 1995; Zeanah, Benoit, Hirshberg, & Barton, 1986). We hypothesized that mothers, who received intervention, will more often have a balanced representation of the attachment relationship with their infants than mothers in the control group. In addition, the first motherhood experiences after the birth of a very preterm infant were studied, as these might affect mother-infant bonding processes (Latva, Korja, Salmelin, Lehtonen, & Tamminen, 2008; Redshaw, 1997).

METHOD

Participants

Two hospitals with level III NICUs and five city hospitals participated in this trial. All infants with a gestational age of <32 weeks and/or a birth weight of <1500 grams, and whose parents were living in Amsterdam were eligible for the study. Exclusion factors were: severe congenital abnormalities, maternal drug use or severe physical or mental illness, non-Dutch speaking families for whom no interpreter could be found, and participation in another trial on post discharge management (for a detailed description see Koldewijn et al., 2009). As language capacity is vital for the interview method used, only mothers who were able to speak and express their feelings in Dutch or English were asked to participate in this part of the study. At 18 months 162 mother-infant dyads were participating in the study. 33 mother-infant dyads were excluded because of poor Dutch or English language and 3 mothers of 3 triplets were excluded. After exclusion 120 mother-infant dyads were eligible. For reasons of feasibility, a random selection of 70% of the mother-infant dyads (n=84) was asked to participate in this part of the study. Finally, 78 mother-infant dyads consented to participate in this part of the study (42 intervention and 37 control group) as 6 mothers refused participation, because they felt too busy.

The intervention

The IBAIP is a post-discharge preventive intervention program for infants at risk from birth to 6 to 8 months and their parents. The goal of the IBAIP is to enhance the infant's social and environmental interactions without distress and to reinforce the infant's motivation and autonomy to explore and to learn from the information. The intervention is guided by the Infant Behavioral Assessment (IBA) (Hedlund & Tatarka, 1988). The IBA is an observational tool based upon the conceptual framework of Heidelise Als (Als, 1986) that underlies the Newborn Individual Developmental Care and Assessment Program (NIDCAP) (Als, 1986). The IBAIP helps the interventionist to sensitize parents to their baby's responses to information, in order to assist parents to support their infant's self-regulatory efforts and to adjust the environment to match the neurobehavioral needs of the infant.

The infants in the intervention group received standard care as well as 6-8 home visits by the interventionists, experienced pediatric physical therapists trained in the IBAIP. Each home-visit lasted approximately one hour. Parents were given detailed information about their infant's development in order to guide the infant's explorations and self-regulatory competence. As the infants matured and their neurobehavioral functioning became increasingly stabilized, parents were encouraged to gradually reduce their co-regulatory support and to enjoy their infants' growing independency.

Infants in the control group received standard care, which consisted of regular visits to the pediatrician in the local outpatient pediatric clinic. In case of disturbances like problems in posture and/or tone in the control infants, the pediatrician was free to refer to a general (non IBA-trained) pediatric physical therapist.

Instruments

Perinatal variables were abstracted from the medical records. Socio-demographic data were obtained from a parent-questionnaire.

Mothers' representations of the relationship with their infants were assessed with the Working Model of the Child Interview (Zeanah et al., 1995). This interview is designed to elicit parents' feelings about their relationships with their children. During the interview the mother is asked about her experiences with and her perceptions of her child. Developed in reference to the Adult Attachment Interview (AAI, Main & Goldwyn, 1982), the WMCI focuses on the parent's emotional reactions during pregnancy, perception of the infant's personality and development, characteristics of the relationship with the infant, reactions to infant behavior and distress, and anticipated difficulties in later development. The parent is also asked to provide specific examples to illustrate the infant's personality and

behavior. The children are not present during the interview. The interviews are videotaped and take about an hour. The interviews are rated with a 5-point Likert scale in six scales that are content free: richness of perception, openness to change, intensity of involvement, coherence, caregiving sensitivity and acceptance. In addition an assessment of the importance of two themes, infant difficulty and fear for safety as content features of the caregiver's representation is made, as well as a rating of the affective tone (joy, anger, anxiety, and indifference) of the caregivers' way of answering to questions. The scores on these features of the narratives by the mothers result in a classification of the mental representations of their children into one of three categories: balanced (secure), disengaged (insecure), and distorted (insecure). *Balanced* representations are characterized by narratives that convey coherence, openness to change, richness of detail, and a sense of the mother as engrossed in her relationship with her infant. These mothers value and enjoy their relationship with their infant and are aware that this relationship affects their child's behavior and development. Mothers who have *disengaged* representations of their children are characterized by emotional distancing from their infant or expressing aversion to the infant. Their narratives show a lack of emotional involvement in the relationship and an unwillingness to change their representations. Mothers with *distorted* representations are more involved and may have a lot to say. However, the descriptions of their infants are incoherent, confused, contradictory, or even bizarre. Mothers with distorted representations may be preoccupied by other concerns, be anxiously overwhelmed by the infant, or may even look to the infant as an adult or buddy. Both disengaged and distorted representations are considered as nonbalanced representations (Benoit, Parker, & Zeanah, 1997).

As a preterm delivery reflects an unexpected, sometimes even traumatic and early ending of pregnancy, further study was done, looking at the content of the answers given by the mothers during the WMCI. Conventional content analysis (Hsieh & Shannon, 2005) was used to gain a richer understanding of the maternal emotions and experiences after very preterm delivery. A number of questions, that might be specifically important in case of preterm delivery and potentially useful in intervention (Latva et al., 2008; Redshaw, 1997) was analyzed in detail: 1) What was your first reaction when you saw your baby?; 2) How would you describe the first few weeks at home with your child?; 3) Are there any experiences which your child has had, that you feel may have been a setback for him/her?; 4) Knowing what you know now, if you could start all over again with your child, what would you do differently?; 5) Do you ever worry about your child? What do you worry about? The answers to these first

two questions were coded into either a generally negative or a positive experience, and the answers to questions 3 and 5 were distinguished into agree or disagree and the elaborations are described.

Procedure

The interview took place at home when the children were 18 months of (corrected) age and was carried out by an experienced psychologist, blinded for group assignment. In case of multiple birth (26%), the mother was interviewed on both children separately. All interviews were carried out by one trained examiner (D.M.). The interviews were subsequently rated by a trained rater, and 25% of the interviews (n=20) was rated by a second trained rater. Interrater percentage of agreement was 80% (Cohen's kappa = 0.56). Interrater differences were resolved by discussion, and the consensus classification was used for data analysis. The Medical Ethics Committee of all hospitals involved in this trial approved the study. This trial is registered with controlled-trials.com, number ISRCTN65502576. All participating parents signed informed consent.

Statistical analyses

Characteristics of the intervention and control group were compared with t-tests and Chi square tests. Differences in maternal WMCI classifications between intervention and control group were analyzed with Chi square test. Analyses of variance and Chi square tests were used to study relations between WMCI classifications (balanced vs. nonbalanced) and neonatal and maternal socio-demographic characteristics. To control for multiple birth, all analyses were also performed for one (randomly selected) mother-child dyad per family.

The results of the conventional content analysis (Hsieh et al., 2005) are described and Chi square test was used to analyze differences in characteristics and representations according to specific positive or negative experiences.

All statistical analyses were performed using version 15.0.1 of the Statistical Package for Social Sciences (SPSS 15.0.1, Chicago, IL, USA). An alpha level of 0.05 was used for all tests of significance.

RESULTS

Tables 1a and 1b show the neonatal and socio-demographic characteristics of the 78 infants and their mothers. Maternal socio-demographic and infant neonatal characteristics were well balanced between the intervention and control group, except for oxygen dependency, which was more in the intervention group.

Table 1a Neonatal characteristics of intervention and control group infants

	Intervention (n=41)	Control (n=37)
Neonatal characteristics		
Gestation (wk), mean (SD)	30.2 (2.0)	30.1 (2.2)
Gestation < 28 weeks	5 (12%)	4 (11%)
Birth Weight (g), mean (SD)	1309 (340)	1331 (363)
Gender infant: male/female (%)	57/44	43/57
Multiple birth (twins), n (%)	10 (24%)	10 (27%)
Oxygen 36 weeks post menstrual age, n (%) †	11 (29%)	3 (9%)
Intraventricular haemorrhage (IVH), n (%)▫	9 (23%)	5 (13%)
IVH grade I +II / III + IV	6/3	3/2
Periventricular leukomalacia (PVL), n (%)▫	3 (7%)	4 (11%)
PVL grade 1 / 2+3	3/0	2/2
Caesarean section, n (%)	25 (61%)	19 (51%)

Note: Numbers are given as number of infants unless otherwise stated
Analyses of variance and Chi-square tests
† Oxygen use at 36 weeks post menstrual age is a marker of chronic lung disease
▫ Intraventricular haemorrhage (IVH) was defined according to Papile et al. (1983),
▫ Periventricular leukomalacia (PVL) according to De Vries et al. (1992)

Table 1b Socio-demographic characteristics of intervention and control group mothers

	Intervention (n=41)	Control (n=37)
Socio-demographic characteristics		
Maternal age, years, mean (SD)	32.5 (5.3)	31.5 (4.6)
Firstborn child	28 (69%)	28 (76%)
Mother born in Netherlands	30 (73%)	26 (70%)
Marital status:		
Single mother	3 (7%)	3 (8%)
Living with a partner	38 (93%)	34 (92%)
Maternal education:		
No high school graduation	11 (27%)	10 (27%)
High school graduation	30 (73%)	27 (73%)

Maternal representations in the intervention and control group

No differences were found in the quality of maternal representations of the relationship with the child in the intervention and control group mothers (Table 2). In both groups about 70% of the mothers had balanced representations and 30% had nonbalanced representations. The number of mothers with disengaged and distorted representations was equally distributed.

Table 2 Maternal WMCI classifications

	Intervention (n=41)	Control (n=37)	Total (n=78)
Balanced, n (%)	29 (71)	25 (67)	54 (69)
Disengaged, n (%)	6 (15)	7 (19)	13 (17)
Distorted, n (%)	6 (15)	5 (13)	11 (14)

Note: Chi-square=0.26, p=0.88

No associations were found between maternal attachment representations and neonatal or socio-demographic characteristics (Table 3). Regarding twins was found that different representation classifications for the children occurred in 10 out of 20 mothers (50%). Analyses with only one of the children did not show any different results than the results in the total group of mothers and children.

Table 3 Relations between neonatal and socio-demographic characteristics with WMCI classifications (balanced vs nonbalanced)

Total group of mothers (n=78)	Balanced (n=54)	Nonbalanced (n=24)	F/X^2
			X^2
Gender infant: male/female (%)	48/52	54/46	0.24
Multiple birth (twins), n (%)	15 (28%)	9 (37%)	3.23
Oxygen 36 weeks post menstrual age, n (%)	10 (18%)	4 (17%)	0.03
Intraventricular haemorrhage (IVH) n (%)	9 (17%)	5 (21%)	0.14
Periventricular leukomalacia (PVL) n (%)	6 (11%)	1 (4%)	1.02
Firstborn child	39 (72%)	17 (71%)	0.02
Mother born in Netherlands	38 (70%)	18 (75%)	0.18
Marital status:			
Single mother	4 (7%)	2 (8%)	
Living with a partner	49 (93%)	22 (92%)	0.46
Maternal education:			
No high school graduate	18 (33%)	3 (12%)	
High school graduate	36 (67%)	21 (88%)	3.66

F=Analyses of variance and X^2=Chi-square tests
**p≤.05*

Content Analysis

The content analysis refers to the total group of 78 mothers (Table 4), because the intervention and control group did not differ in their answers to any of the five specific questions.

Table 4 Content analysis WMCI

	Total group of mothers (n=78)
1. What was your first reaction when you saw the baby?	
Negative (distance, unreal, not in love, unfamiliar)	39 (50%)
Positive (relief, happy, beautiful)	39 (50%)
2. How would you describe the first few weeks at home?	
Negative (scared, strange, difficult, hectic, isolated)	29 (37%)
Positive (happy, quiet, special)	23 (29%)
Both negative and positive	22 (28%)
3. Are there any experiences which your child has had, which you feel may have been a setback for him/her?	
Yes	51 (66%)
No	27 (34%)
What do you think has been a setback?	
Prematurity (and infant's start)	44 (56%)
Other (missing father, medical problems, vacation without child)	33 (44%)
4. Knowing what you know now, if you started all over again with your child, would you do things differently?	
Yes	34 (43%)
No	44 (57%)
What would you do differently?	
I would be more careful during pregnancy	19 (24%)
I would not have become pregnant	14 (18%)
Child rearing (less over protecting, more strict)	14 (18%)
I would be there for my child more often	12 (15%)
I would seek help for myself earlier	9 (12%)
5. Do you ever worry about your child?	
No	27 (34%)
Yes	51 (66%)
What do you worry about?	
Health	30 (39%)
Development	22 (28%)
Health & Development	11 (14%)
Character	9 (12%)
Attention Deficit Hyperactivity Disorder (ADHD)	5 (6%)

First reaction to the infant

Fifty percent of the mothers reported negative emotions concerning the first time they saw their baby, like fear about how small the infant was and its bio-medical condition, and they expressed feelings of alienation. Of these mothers 31% felt the baby was not theirs. Negative feelings and feelings of distance towards their child were not influenced by the way of delivery, like a Caesarean section (X^2=2.62, p=0.10). Mothers with negative first experiences more often were found to have nonbalanced representations (X^2=6.26, p=0.04).

The first two weeks at home with the child

Twenty-nine percent of the mothers presented overall positive experiences in the first two weeks at home with their infant; 37% of the mothers reported negative experiences. In most cases those negative feelings consisted of feelings of fear, stress and worries. 28% of the mothers felt ambivalent about the first two weeks at home, both negative and positive. The recollections of the first experiences are found to be associated with the maternal representations of their child; mothers who presented negative experiences in the first few weeks at home with their infant more often had nonbalanced representations (X^2=10.27, p=0.01).

Setback for the infant

Almost two third of the mothers (66%) reported that their infant experienced a setback. Especially the preterm delivery and the hospital admission period are considered as setbacks (56%). No relation in experiencing setbacks for the child and the quality of the maternal attachment representations was found (X^2=2.45, p=0.12).

Starting all over, doing things differently

43% of the mothers would do things differently if they could start all over again with their child. Things they would do differently are: being more careful during pregnancy (24%); not getting pregnant at all (18%); child rearing issues (18%); being there for the child more often (15%); and getting help for themselves earlier on (12%).

The fact that mothers wanted to do things differently if they could start all over again tended to be associated with more nonbalanced representations (X^2=2.88, p=0.09). An affirmative answer to this question ('Yes, I would do things differently') was related to the affective tone subscale 'Guilt' (F(1, 68)=17,74, p=0.00), illustrating the guilt ridden connotation of the given answers.

Worries about the child

66% of the mothers had worries about their child. The most important concerns were the physical health and growth of the child (39%) and the child's development (28%). These were often mentioned together by the same mother (14%). Some mothers worried about their child's character (12%) or Attention Deficit Hyperactivity Disorder (6%).

The content of the worries was not related to the quality of the maternal attachment representations or the intervention (X^2=2.51, p=0.28). However, 8 of the 9 mothers who were concerned about their child's character had nonbalanced representations.

DISCUSSION

No differences were found in maternal attachment representations between mothers that received IBAIP intervention and control group mothers. In both the intervention and control group about 70% of the mothers had balanced representations and 30% had nonbalanced representations. The amount of disengaged and distorted representations was equally distributed. These percentages are different from one other study using the WMCI in Italian mothers of preterm born children at 18 months corrected age (Borghini et al., 2006): in this report the maternal representations were 30% balanced, 32% disengaged, and 38% distorted. Our distribution is similar to that found in studies of mothers of healthy term born children (Borghini et al., 2006; Rosenblum, Zeanah, McDonough, & Muzik, 2004; Wood, Hargreaves, & Marks, 2004). Borghini et al. (2006) did find that mothers of very preterm children had more non-balanced WMCI classifications at 6 months than at 18 months post term, hence the time when the interview is done, may be important and mothers of very preterm infants may need more time to develop a balanced attachment representation.

The quality of the maternal attachment representations was not related to infant health status at birth, as was found in an earlier study on attachment in mothers of very preterm children, that used the AAI (Brisch et al., 2005). The socio-demographic characteristics of the mothers, like single parenthood, educational level or ethnicity, were also not related to the attachment representations.

Although the distribution of balanced versus nonbalanced representation was similar to that found in studies on mothers and term born infants, it may be especially important for children at risk and their mothers to develop secure attachment relationships. Important information that may provide clues to improve caretaking of mothers of very preterm infants emerged

from the content analyses of the interview concerning the experience of specific circumstances surrounding very preterm delivery. Half of the mothers of very preterm infants felt they had to cope with negative feelings when first seeing their baby and 65% had negative or ambivalent feelings in the first weeks at home with their baby after hospital discharge. A nonbalanced representation was found to be more often associated with reports of such negative feelings. These results suggest that mothers of very preterm infants may need additional support in coping with negative feelings surrounding these early experiences. Maternal support could start immediately after birth to promote initial moments of positive interaction between mother and infant. Direct physical contact between mother and infant could be promoted even more strongly immediately after birth. If this is not possible, the mother may need extra support during and after the first moments of contact with her baby in the hospital. In a study on maternal recollection of experiences after preterm delivery the importance of the first contact between mother and infant was also emphasized (Latva et al., 2008; Redshaw, 1997). The staff, especially the nurses and psychologists working in neonatal units could play an important role in this regard. In a recent meta-synthesis several recommendations for neonatal nursing, like guided participation in caretaking were proposed, that could be useful in this regard (Aagaard & Hall, 2008).

More than half (56%) of the mothers experienced the preterm delivery as a setback. Their concern about the child's health and development may be reduced by providing (tailored) information about the possible consequences of preterm delivery, both the chances for developmental problems, as well as the fact that many children develop without severe disabilities. This may also prevent both psychological distress and overprotective caretaking in mothers due to the often perceived vulnerability of the child (Allen et al., 2004). A lot of mothers deal with feelings of guilt towards their child and 43% of the mothers report in our study that they would do things differently if they could start all over again. Feelings of guilt combined with anxiety could negatively interfere with mother-infant interaction. Easily accessible psychological support, both before and after hospital discharge, in order to give room for the mothers to talk about their fears, worries and feelings of guilt, might help mothers of very preterm born children in building a healthy and positive relationship with their child without any restraints.

Clinical implications

This study shows that the IBAIP in its current form did not affect maternal attachment representations. Given the importance of the early mother-infant relationship for infant development, and the fact that 30% of the mothers of very preterm infants had nonbalanced attachment representations, early (psychological) support for the mothers is recommended. Especially the first negative experiences of the mothers in the hospital and in the home situation may be a risk factor for a positive and healthy relationship. Therefore, early support should address the mothers' feelings during the first contacts with her baby as well as her first experiences with the child at home.

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6

Discussion

'Ik heb het contact en de werkwijze van onze IBAIP- begeleidster als zeer prettig ervaren en nog steeds. Als ik onzeker was in het omgaan met mijn baby kon zij mij heel goed steunen en op het goede pad houden. Ze gaf me ook het gevoel dat ik het als ouder goed weet en de juiste basis heb om mijn baby te helpen. Ze is erg goed geweest voor mijn zelfvertrouwen en leerde me steeds goed naar hem te kijken en naar wat goed voor hem is.'

DISCUSSION

In this thesis the effect of the IBA Intervention Program for very preterm born infants and their parents was evaluated in relation to mother-infant interaction, maternal psychological well being, maternal parenting stress and maternal attachment and bonding processes. A high prevalence of morbidity in different developmental domains has been reported for very preterm infants. In addition, the rate of preterm births in the Western world is still increasing due to improvements in obstetrical care, fertility treatments, and the advanced maternal age of pregnant women. Hence, effective intervention programs to reduce the developmental morbidity in very preterm infants are needed, both for the children and their families. Recent meta-analyses show that early post-discharge interventions have the highest impact on long-term infant cognitive development (Spittle, Orton, Doyle, & Boyd, 2007; Vanderveen, Bassler, Robertson, & Kirpalani, 2009). However, only few studies have focused on the impact on mother-infant interaction and parental outcome, next to long term developmental outcome in children.

The IBAIP is based on the assumption that the parent's availability and adequate responsiveness strengthens the infant's regulatory competence and development. Self regulatory competence is the ability to modulate emotion, self-soothe, delay gratification, and tolerate change in the environment. This affects the infants' ability to explore and interact with their environment and consequently their learning processes. Very preterm infants have been found to show greater difficulty in self regulation behaviors than term infants (Clark, Woodward, Horwood, & Moor, 2008; DeGangi, 2000). IBAIP guides parents in supporting their infants' self-regulatory competence in 6-8 sessions. Consequently the infants may learn more easily how to adjust to their environment and how to explore new information. The parents learn how to respond sensitively and effectively to their infants' needs.

A multicenter randomized trial was conducted to evaluate the effect of IBAIP on infant development. Important improvements were found in the infants' mental and motor development over the first two years (Koldewijn et al., 2009; Koldewijn et al., in press). It was hypothesized that the IBAIP, next to the long term developmental improvements in the children, may also result in positive effects on the parents and the parent-infant interaction. In addition the IBAIP might influence the mothers' feelings of psychological well being and parenting stress, as well as the processes of bonding and attachment, because of the affected interaction processes.

IBAIP and mother-infant interaction

The first hypothesis was that IBAIP would increase positive and sensitive maternal behavior and improved self-regulated infant behavior during mother-infant interaction. Mother-infant interaction was measured with the Still-face procedure. This procedure was chosen because it is a standardized procedure to observe both normal face-to-face parent-infant interaction and the infants' reactions to a sudden interruption of the interaction. It needs a limited amount of time and provides insight in the infants' frustration experience and regulatory responsiveness.

The hypothesis was partly confirmed. More sensitive and less intrusive interaction behavior in the mothers was found with the Still-face procedure at the infant's corrected age of 6 months. A small increase in positive maternal interaction behavior was found during the two minutes lasting first interaction episode, which reflects baseline interactive behavior. No group differences were seen during the reunion episode, after the slightly stressful Still-face episode. Better regulated infant interaction behavior and less stress behavior was not found in the intervention infants. It may not have occurred, or the duration of the procedure (in total six minutes) could have been too short for meaningful differences to appear. The kind of coding procedure of the Still-face procedure may also not have been appropriate, as the ICEP does not allow subtle distinctions in self-regulatory behavior (see also further in the strengths and limitations section of this study).

Subtle intervention effects were found in the intervention infants regarding more environment focused behavior during the interaction episodes and less positive interaction behavior (smiling). At about 6 months of age term infants become increasingly interested in their environment (Legerstee, Pomerleau, Malcuit, & Feider, 1987) and in objects (Tronick & Cohn, 1989). More environment-focused interaction behavior in the intervention group infants might therefore be related to advanced development. This observation is supported by the finding that the infants receiving intervention were more advanced on the Bayley Scales of Infant Development compared to the control group infants at 6 months of age (Koldewijn et al., 2009). Another interpretation of the differences in the infant interaction behaviors could be that the intervention infants showed less affection and actively avoided their mothers, as they smiled less and looked at their mothers less frequently. The increase in positive engagement behavior of the mothers then could be interpreted as more intrusive or as compensating for a less engaged infant. However, this possibility seems less plausible, as positive engagement behavior of the mothers was associated with higher ratings in sensitivity and less

overcontrolling behavior. Unfortunately, the observation of the interaction did not allow a further distinction in infant's gaze aversion, as either self regulatory behavior following emotional arousal or as real object or environment focused behavior. Infants can use gaze aversion as a self-regulatory coping strategy to prevent themselves from getting over aroused or to focus their attention deliberately on an object of interest in the environment.

In general, preterm infants' interaction behavior is less active and clear, compared to term infant interaction behavior (Eckerman, Hsu, Molitor, Leung, & Goldstein, 1999). This makes it for mothers of preterm infants more difficult to interact with their infants. Although, the intervention infants showed less positive interaction behavior than the control group infants, their mothers were more sensitive and less intrusive. This shows that the IBAIP improved mothers' ability to read their infant's behavior better and to respond in an appropriate way. Sensitive maternal interaction behavior is important for prevention of emotional and behavioral problems later in life (Denham et al., 2000; Forcada-Guex, Pierrehumbert, Borghini, Moessinger, & Muller-Nix, 2006) and it is an important predictor of infant neurobehavioral development and infant attachment status (Feldman & Eidelman, 2003; Goldberg & DiVitto, 1995). The IBAIP resulted in increased sensitive mother-infant interaction and may therefore have contributed to the prevention of emotional, behavioral and neurodevelopmental problems and insecure infant attachment status.

The Still-face paradigm has not been used often to assess mother-infant interaction after very preterm delivery. Therefore, it was interesting to see that very preterm infants (both intervention and control group) showed the expected Still-face procedure behaviour. During the episode in which their mothers held a blank face they looked around more than in the first episode and they smiled less, while checking frequently whether their mothers had already changed their behavior. Also a carry-over effect of reduced looking and increased negative affect from the Still-face to the subsequent reunion period was observed (Adamson & Frick, 2003; Weinberg & Tronick, 1996). So preterm infants also noticed the change in their mothers' responses. Limited occurrences of emotions, either positive (6%) or negative (5%) were seen during the actual interaction episodes with their mother (normal play and reunion episodes). This sensitivity of preterm infants to the Still-face procedure has also been recently reported in other studies (Erickson & Lowe, 2008; Hsu & Jeng, 2008; Montirosso, Borgatti, Trojan, Zanini, & Tronick, 2008; Segal et al., 1995). Nonetheless there are differences in the reactions of the preterm children in the different studies published. Some studies suggest that preterm infants do not react

differently compared to full term infants. Segal et al. (1995) reported that 7 month-old black preterm infants (gestation range 26-32 weeks) smiled less during the Still-face procedure compared to term infants and that both preterm and term infants did not show the expected increase of negative behavior. Montirosso et al. (2008) found no differences between 6 to 10 month-old Italian preterm (< 37 weeks of gestation) and full term infants, but they did find a difference in self regulatory behaviors as reflected in more distancing behavior in preterm infants. We did not observe any distancing behavior, e.g., turning and twisting away from the parent by rotating the shoulders and trunk without engaging an object in our infants. Hsu & Jeng (2008) compared Taiwanese 2 month's old preterm (gestation range 24-34 weeks) and term infants during the Still-face procedure and they found longer duration of negative affect in preterm infants and shorter latency to negative states. These findings partially match our results. In our study all 112 infants were born very preterm (< 32 weeks). Our infants also showed little smiling and negative affect for about 5 percent of the time during the reunion episode, which was less than in the study of Hsu et al. (2008). Thus it seems that (very) preterm infants do show the expected Still-face behaviour. Future studies on interaction and self regulation capacities are necessary to compare infants with different gestational ages concerning their negative affect regulation capacities and their amount of positive affect during mother-infant interaction.

We also observed gender differences in the infants' reactions. Boys looked and smiled more at their mothers during the interaction episodes than girls. These differences were also seen in some other studies on the Still-face procedure with term infants (Tronick et al., 1989; Weinberg, Tronick, Cohn, & Olson, 1998). In those studies boys displayed more positive as well as negative affect, focused more on the mother and displayed more signals expressing escape and distress, or demands for contact. On the other hand, girls showed more interest in objects. The fact that preterm infants show similar gender differences in interaction behavior as term infants, adds to the information on preterm interaction behavior. It also emphasizes the importance of the individualized approach of the IBAIP in which neurobehavioral strategies are continuously assessed and modified depending upon the infant's behavior within the infant's zone of proximal development.

The amount of positive affect in our preterm infants was less than observed in earlier studies and was also less than seen in term infants. These differences may be related to the infants' preterm status or to the mothers' behavior. The total group of mothers in our study showed positive interaction behavior in about 30% of the time. In studies of face-to-face

interactions between mothers and healthy term infants, expressions of positive affect occurred in about 42% of the time in the mother and in about 15% of the time in the infant (Cohn & Tronick, 1987; Tronick et al., 1989). The amount of positive affect shared by both the mother and the preterm infant therefore seems to be smaller in our study group compared to mothers and their healthy term infants. The explanation might be that the mothers' lower level of positive affect resulted in less positive affect in their infants or mothers' lower level of positive affect may have been generated by the infants' lower levels. Whatever the initial cause, as a dyad these preterm dyads seem to be less positive than full term dyads, regardless of intervention. Mothers of very preterm infants have to work harder to initiate and maintain interaction with their infants, as they receive little positive responses from their children. The fact that very preterm infants are in general less self-regulated and more easily over-aroused, which is sometimes expressed by more negative affect, also complicates mother-infant interaction, especially mother-infant synchrony and the repair of mismatched states (Feldman & Eidelman, 2007; Tronick, 2007). When trying to improve the quality of mother-infant interaction in preterm dyads such difficulties need to be recognized.

IBAIP and maternal psychological distress

Our second hypothesis was that the IBAIP might reduce maternal psychological distress as a result of the support provided in the home situation by professional IBAIP trained interventionists. We studied this aspect with the General Health Questionnaire (GHQ). No differences in psychological distress between the intervention and control group were found after IBAIP at 6, 12 and 24 months (corrected age). This makes our previous results on mother-infant interaction (Still-face procedure) even more intriguing. Although the mothers in the intervention group felt as psychologically distressed as in the control group, they were able to interact more sensitively with their infants during the Still-face procedure.

Most early intervention programs do not report on maternal psychological well being but focus on the child's development and behavior. They are mostly conducted during the child's hospital stay and the results are conflicting (Hagan, Evans, & Pope, 2004; Preyde & Ardal, 2003).

The Infant Health and Development Program (IHDP) (Klebanov, Brooks-Gunn, & McCormick, 2001), however, reported less emotional distress in mothers after intervention, but this intervention program consisted of a home visiting program during the first 3 years, while our intervention program consisted of only 6-8 home visits during the first 6 months after discharge from hospital. Another difference between the IBAIP and the

IHDP is, that the IHDP aims primarily at reducing emotional distress in the mothers, whereas the IBAIP primarily aims at improving the infant's development. It is therefore not surprising that the IBAIP did not result in reducing psychological distress in the mothers. Moreover, the IBAIP was executed by pediatric physical therapists who are not specifically trained in diagnosing and/or treating psychological distress in mothers. Another early intervention program was recently reported (Doyle et al., 2009). This program consisted of 9 home visits over the first year by both a physical therapist and a psychologist. The focus of the program was on the parent-infant relationship, parental mental health and infant development, and it showed less anxiety and depression in mothers in the intervention group at 2 years (corrected age of the infants). It may be that extra psychological support may have caused this better maternal psychological outcome.
Because of the high amount of both the intervention and control group mothers with clinically relevant psychological distress symptoms at 6 months (corrected) age we compared our results with mothers of term infants. The underlying assumption was that in general becoming a mother requires psychological adjustment. The significantly higher rate of clinical cases of distress in the mothers of very preterm infants showed that mothering a term infant, is not as distressing as mothering a very preterm infant. However, two years after birth the psychological stress levels in both the intervention and control group mothers have been normalized compared to norm reference women. Although, 30-50% of the children had abnormal developmental scores at 24 months (corrected age) (Koldewijn et al., in press), no elevated levels of stress were found at this age. This might be explained by parents' tendencies to set appropriate expectations for their children, and to adapt to and compensate for their child's needs (Greenberg & Crnic, 1988; Taylor, Klein, Minich, & Hack, 2001). However, we have to keep in mind that the respondents to the questionnaires were higher educated and more of them were Dutch compared to the non-respondents. This may have resulted in an underestimation of psychological distress at 24 months of age, as higher levels of stress in lower educated mothers have been reported (Davis, Edwards, Mohay, & Wollin, 2003; Taylor et al., 2001).

IBAIP and maternal parenting stress

The third hypothesis studied was that the IBAIP might reduce maternal parenting stress as a result of the support at home by the professional interventionists. One of the key components of the IBAIP is to support parents in raising their child in such a way that the parents feel confident and positive about themselves and their child. Maternal parenting stress was measured

at 12 and 24 months with the Parenting Stress Index. The IBAIP was found not to influence maternal parenting stress at 12 months. However, at 24 months (corrected age of the infants) maternal parenting stress differed in some aspects between the intervention and control group; intervention group mothers assessed their children as happier and less hyperactive and distracted. These results are similar to the Norwegian study on a modified version of the Mother Infant Transaction Program (Kaaresen et al., 2008), that also showed less parenting stress in their intervention group at 24 months on the mood and distractibility/hyperactivity subscales. However, in our study the intervention group mothers reported more social isolation after 24 months. Maybe the intervention has increased the parents' availability and feelings of responsibility towards their child, which may have resulted in less parental social activities. More information from the parents is needed to detect which aspects of the IBAIP have contributed to there feelings of social isolation. Specific evaluation in which parents can express their emotions and reasons for social isolation can be useful in a possible extension of the IBAIP. Moreover the IBAIP interventionist should be aware of such a possible side-effect of the intervention.
Weak to moderate correlations were found between maternal parenting stress and maternal psychological distress at 12 and 24 months as shown in Chapter 4. Hence, when assisting parents in coping with the consequences of a preterm birth, it seems relevant not only to pay attention to aspects of parenting, but also to emotions and feelings of stress of the parents themselves.
Parenting a very preterm infant, even a very high risk infant, was not found to lead to high levels of parental and parenting stress 1-2 years after birth, which was also found in an other Dutch study (Pal et al., 2008). Unfortunately, maternal parenting stress was not measured 6 months after birth as was done for maternal psychological distress, which could have provided a more complete insight in the different aspects of maternal stress over time. Hence it is possible that maternal parenting stress levels were elevated at 6 months and normalized after 12 months.

IBAIP and maternal attachment representations

The fourth hypothesis studied was that IBAIP support might have a positive impact on the mother-child relationship, reflected in the maternal attachment representations regarding her child and her relationship with her child. To investigate this, the mothers were interviewed with the Working Model of the Child Interview at 18 months corrected age of their child. We chose 18 months corrected age to assess maternal attachment representations, because we assumed that the mother-infant bonding

process is then in most cases developed. By that time the critical period for most of these infants and their parents is over and mothers might better be able to reflect on their experiences with their very preterm child.
No differences were found between maternal attachment representations of mothers from the intervention and control group. Although, we speculated that the extra support given to the parents during the first vulnerable months with their child at home could have been beneficial for the mother-child relationship as well, it is important to realize that the IBAIP was not focused on an improvement of maternal attachment representations.
To the best of our knowledge no post-discharge intervention program aimed at the improvement of infant developmental outcome, has been evaluated in regard to maternal attachment representations. Hence our results cannot be compared to other studies.
The distribution of balanced and non-balanced attachment representations in the total group of mothers was 70% vs. 30%, respectively. Only two other studies using the WMCI assessing maternal attachment representations in mothers of very preterm born infants have been reported (Borghini et al., 2006; Korja et al., 2009). Borghini et al. (2006) showed less balanced attachment representations at 18 months of the infant's corrected age. This might be explained by a difference in socio-economic background between our studies. In the Borghini et al. (2006) study, the level of socio-economic status of the families was lower in the group of preterm infants than in that of term infants. Lower socio-economic status increases the risk for problems in the mother-infant relationship (Wille, 1991). The study of Korja et al. (2009) showed results more comparable to our results; they found balanced attachment representations in 55% of the mothers of very preterm infants and no differences with a group of mothers of term infants at the infant's age of 12 months. It seems that, in spite of concerns regarding the mother-infant relationship due to very preterm birth, most mothers are able to establish a secure bonding with their preterm born child. More in depth content analysis of some of the answers given by the mothers in our study showed that especially the first contact moments (soon after delivery) between mother and child in the hospital and in the first few weeks at home are important for the developing mother-child relationship. Mothers with more negative memories about their first moments of contact with their infant either in the hospital or in the first weeks at home showed more often nonbalanced representations. Therefore it seems to be very important to support mothers early, both during the infant's stay in the hospital and after hospital discharge at home in order to build a healthy attachment relationship.

Strengths and limitations of the study

The strength of our study is that all group comparisons were done within the design of a randomized controlled trial (RCT), which allows the assumption that important characteristics and capacities of mothers and infants were equally divided between the groups at the start of the intervention procedure. Despite the randomization procedure, however, the groups were found to differ in some neonatal characteristics. These could be statistically controlled.

A baseline measure of all outcome variables used in this study (i.e., pretest-posttest design) in addition to the RCT might even have been better to show if any significant improvements over time had occurred more clearly in the intervention group. However, this was not feasible, partly due to the fact that the preterm birth was often unexpected and no time remained to gain baseline information (i.e., on psychological distress). Also ethical reasons were important, as the collection of such data could have been a considerable burden to the parents in a very emotional period in their lives. A design with a dummy-treated control group, that controlled for potential attention effects of the intervention, implying home visits to families with preterm born infants that did not address the infants' development or maternal well being, was also not considered feasible both for conceptual as well as practical reasons. Offering an intervention to parents consisting of only parental attention is difficult to design in an objective manner. Attention can vary from social talk to giving compliments and a standardized way of giving attention seems not realistic. Practically, it was not feasible, because of the costs and time investment for the interventionists to visit the families 6-8 times. Consequently, we cannot determine which part of the intervention procedure (only attention, or the actual information and support provided) exactly is responsible for the results. However, it seems unlikely that the positive outcomes in this study would be due to attention to parents only, especially in view of the lack of effects found on maternal psychological distress and parenting stress.

A limitation in this study is the disproportionate attrition regarding the questionnaire response rate from our original sample of families of lower educated mothers and mothers born outside the Netherlands. Such difficulties have also been reported in other long-term follow-up studies (Wolke, Ratschinski, Ohrt, & Riegel, 1994). However, the possibility that this has contributed to a positive bias of our results seems small, as a low education or not being born in The Netherlands was not found to be related to maternal psychological distress. If such variables did show relationships to the outcome the analyses were adjusted for these variables, for instance for parenting stress. Furthermore, the number of respondents

to the questionnaires at 24 months turned out to be lower in the control group than in the intervention group. This may indicate an intervention effect in itself, as intervention mothers might have felt more dedicated to the study and therefore cooperated better. Unfortunately, high drop out rates at 24 months reduce the power of the results and require a stronger effect size, before the effects appear in the analyses.
A further limitation is that very little information from the fathers was collected. The aim of the study was to evaluate the effects of the IBAIP on the parent-infant interaction, parental and parenting stress, and the parental attachment representations. We asked the parent spending the most time with the child to respond to the questionnaires and to participate in the Still-face procedure and the Working Model of the Child Interview. At all times the majority of the participants/responders consisted of mothers. Therefore, no or very little information is collected from the fathers. It is therefore unclear whether or how the IBAIP might have affected the father-infant interaction, paternal attachment representations and stress. It also remains unclear how the fathers might have contributed to the maternal outcome in this study.

The choice of instruments in this study can also be disputed. The Still-face procedure was chosen because it is a standardized procedure to observe parent-infant interaction that needs a limited amount of time and also evokes stress responsiveness in infants. However, the Infant and Caregiver Engagement Phases (ICEP) system that was used to code the different interaction behaviors does not distinguish between very subtle interaction behaviors. The ICEP discriminates only two self-regulatory behaviors, whereas the IBA (Appendix III) distinguishes 42 self-regulatory behaviors. The IBA's validity and reliability are currently investigated (Koldewijn et al., in preparation).
The General Health Questionnaire (GHQ) provided insight in the psychological complaints of the mothers. However, the GHQ is not often used in populations of mothers after very preterm birth and that limited the possibilities to compare our results with other studies. In order to find out whether the high amount of mothers with clinical levels of psychological distress at 6 months was due to mothering a premature child, we collected GHQ data from a sample of mothers of healthy term born babies. Results showed that mothering a healthy term child does not lead to similar levels of clinical psychological distress. This indicates that the GHQ can be a useful screening instrument to detect clinical levels of psychological distress in mothers of preterm and term infants. However, it does not screen specifically for post traumatic stress disorder, which has been reported to

be a frequently occurring disorder during the first year after very preterm birth (Kersting et al., 2004; Kersting et al., 2009).
The WMCI was chosen because it provided an opportunity for mothers to tell in their own words about their child and their relationship with their child. It also allowed them to emphasize important experiences, in stead of having to answer limited and standardized questions. The WMCI provided insight in the quality of the attachment representations of the mothers, coded by an objective trained coder. In addition more in-depth information became available about the impact of preterm birth on the mother-infant relationship. Although, the WMCI has been used in two other studies with mothers of very preterm babies, no previous study has studied the content of the answers and the relation to the attachment representation.

Clinical implications
IBAIP intervention shows an improvement in sensitive maternal interaction behavior during interaction with their very preterm infant, and a decrease in some aspects of parenting stress, but no reduction in maternal psychological stress and no improvement in attachment representation. The outcome on infant development and behavior (Koldewijn et al., 2009) indicates that IBAIP improves mental, motor and behavioral development at 6 months (corrected age), and this effect was sustained in motor development at 24 months corrected age (Koldewijn et al., in press). Especially, children with bronchopulmonary dysplasia (BPD) and multiple risk factors (BPD and low maternal education / abnormal ultrasound) seem to profit from the IBAIP, as they also showed an improvement in mental development at 24 months corrected age. Mothers of these very vulnerable children might also have profited more from the IBAIP but our results did not show any correlations between BPD or other medical background variables and maternal outcome.
The results suggest that implementation of the IBAIP as standard care in the Netherlands will benefit many preterm children, especially the more vulnerable children. However, the IBAIP in its current form needs to be extended with extra support for the mothers as early and extra support during the first months after preterm delivery may prevent clinical levels of psychological distress and a non-balanced attachment representation. Further study should determine what this support should look like and when would be the best time to start this support. Recent results from an Australian early intervention program performed by both a physical therapist and a psychologist showed positive results for maternal psychological well being after very preterm birth at 2 years of age (Doyle et al., 2009). More

information on the content of this early intervention program is needed to determine whether elements of the program could be added to the IBAIP. An early screening instrument for psychological distress, like the GHQ or evaluating the amount of post traumatic stress disorder (PTSD) symptoms, might be helpful in determining which mothers need more extensive support and, if necessary therapeutic intervention, like Eye-Movement Desensitization and Reprocessing or cognitive behavioral therapy (Cloitre, 2009). Mothers without specific PTSD symptoms may benefit from other forms of support, like mindfulness (Kabat-Zinn et al., 1998) or group (counseling) sessions with debriefing elements (Rowan, Bick, & Bastos, 2007) and psycho-educational aspects to cope with the impact of very preterm birth by sharing their experiences with other parents. Another possibility might be to start the support and psycho-education already before the expected premature birth. However, most premature deliveries are unexpected.

The impact of (early) maternal psychological distress on infant behavior and development has been demonstrated in many studies and the impact seems to increase, the older the children get (Miceli et al., 2000; Murray, Fiori-Cowley, Hooper, & Cooper, 1996). Adequate and available psychological care in the NICU for parental support therefore is essential.

We should also take into account that the current IBAIP may have an unexpected side-effect, as mothers in the intervention group reported more social isolation two years after birth of their child. This finding should be taken seriously and deserves further study. An optimal balance between care taking activities and parental well being should be strived for.

When evaluating the effect of the program, the experiences of parents with the IBAIP are important, next to the results based on standardized questionnaires. In this context, parents were asked to complete an evaluation form after the intervention was completed (Appendix IV). The response rate was 72%. Parents assessed the IBAIP as pleasant (92%) and supportive (87%). Most parents experienced positive effects of the intervention in their babies (83%), and 90% of the parents would recommend the intervention to other parents. Although, no positive effects on maternal psychological distress at 6 months were found by using a standardized questionnaire, the results of the parental evaluation showed that parents felt supported by the IBAIP. It seems worthwhile to further investigate the IBAIP effects on parents with more in depth methods, like interviews, in order to find out what parents may have missed in the current program and if and how they would have appreciated extra maternal support.

As a result of the positive outcomes of the present study, especially regarding the improved quality of the mother-infant interaction and the

improvement in infant developmental outcome (Koldewijn et al., 2009), the IBAIP will be implemented as standard care in the Amsterdam region. The current intervention program will be extended with specific maternal support to prevent clinical levels of psychological distress and to promote a secure mother-infant bonding. The extended IBAIP and the quality of care will be evaluated, together with customer satisfaction measurements.

Future research

The unexpected outcome of more frequently reported feelings of social isolation in the intervention group mothers deserves further study. It is necessary to detect what is responsible for these feelings of social isolation and what it actually means for mothers.

Future research should also focus on collecting information on the effects of the IBAIP on fathers. Perhaps the fathers mediated the positive effects of the IBAIP on the improved developmental outcome in the children through their supportive role and their interacting style with their children (Affleck, Tennen, & Rowe, 1991; O'Brien, Heron Asay, & McCluskey-Fawcett, 1999). Furthermore, it would be interesting to interview the fathers with the Working Model of the Child Interview, as it has never been done before with fathers of very preterm infants in order to find out whether very preterm birth affects their attachment representations and if the IBAIP has influenced them. Social and partner support was also not studied during the first 12 months. At 24 months this was only studied in short with the NOSI. Especially during the first year in which the mothers reported high amounts of psychological distress, the role of social and partner support may be important because it could play an important role in the process of mother-infant bonding (Cassidy, 2000; Theran, Levendosky, Bogat, & Huth-Bocks, 2005) and the amount of experienced psychological distress felt by the mother (Affleck et al., 1991; Candelaria, O'Connel, & Teti, 2006). Finally, it seems worthwhile to follow the mothers and children for a longer period of time. The quality of early mother-infant interaction and the high amount of psychological distress in the first 6 months after birth may have consequences for later child developmental and behavioral outcome. Some intervention effects, like improved cognitive developmental outcome, may even be delayed, as was shown in other intervention studies (Achenbach, Howell, Aoki, & Rauh, 1993; McAnulty et al., 2009). A recent study in the Netherlands suggested that very preterm infants might benefit from both sensitive and directive maternal interaction behavior (Weijer-Bergsma, 2009); a structured approach by mothers (i.e., directiveness) seemed to facilitate the development of executive functioning in preterm infants between 7 and 14 months corrected age. Weijer-Bergsma (2009)

suggested that mothers may attempt to compensate for a lack of initiative and activity of their infant by providing more structure. It would therefore, be interesting to see if and how our results are related to later developmental outcome, i.e., executive functioning. Hence follow-up examinations at 3,5 and 5,5 years for all children of the study are currently performed.

Conclusion

IBAIP was found to increase sensitive mother-infant interaction and to decrease some aspects of parenting stress two years after birth. Implementation of the IBAIP is justified, but the current intervention program needs to be extended with specific maternal support to prevent clinical levels of psychological distress and to promote a secure mother-infant bonding.

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SUMMARY

Each year in the Netherlands approximately 8% of all births are very preterm. Very preterm birth has consequences for both the child and the parents. Preterm infants are at increased risk for neurodevelopmental impairments in childhood as a result of neonatal brain injury as well as interruption of the normal process of brain maturation that occurs during the last trimester of pregnancy. Next to and often due to medical complications, preterm infants experience difficulties in their self-regulation, referring to the ability to modulate emotion, self-soothe, delay gratification, and tolerate change in the environment. This affects their ability to explore and interact with their environment and consequently their learning processes.
For parents preterm birth interrupts the normal process of adjustment to parenthood, not only because of the unexpected nature of the birth, but also because of the infant's fragile medical condition and extended hospitalization. Coping with preterm birth can be a difficult and distressing experience for parents and it may lead to post-traumatic stress symptoms. Preterm birth can also complicate the development of a healthy mother-infant relationship. Both the NICU surrounding, in which the physical closeness and caretaking between mother and child is complicated, and the emotional imbalance of the mother may prevent the mother from bonding with her child. Also the mother-infant interaction after preterm birth may be more difficult because of the unclear behavioral signs of the baby, as well as the emotional state of the mother influencing her possibilities to interact sensitively with her baby.
Considering the fact that developmental, social-emotional and health problems of very preterm infants persist even into early adulthood, various types of early intervention programs have aimed to improve the long-term outcome of these children. Up until now only some long term positive effects are found on cognitive developmental outcome by a few post-discharge intervention programs. However, rarely effects of such intervention programs on feelings and functioning of the parents were studied.

In this thesis, the effects of the Infant Behavioral Assessment and Intervention Program (IBAIP) were evaluated. IBAIP is a post-discharge preventive intervention program and aims at early self-regulatory support, as one of the basic elements underlying the infant's development and parent-infant relationship and interaction. **(Chapter 1)**
The results of IBAIP were studied using a randomized controlled trial in a sample of very preterm born infants and their parents. Attention was paid

to mother-infant interaction, maternal psychological well-being, maternal parenting stress and maternal attachment representations.
In **Chapter 2** the effect of the IBAIP on the mother-infant interaction is described. Mother-infant interaction was assessed with the Still-face procedure in which 112 mother-infant dyads participated, 57 from the intervention group and 55 from the control group. The Still-face procedure consists of three episodes. In the first 2-minute episode the mother is instructed to interact with her child as she normally does in the home situation. The second 2-minute episode is the Still-face episode in which the mother is not allowed to interact with her child and to keep a blank face. During the third episode the mother is allowed to interact again with her child. Micro-analytic coding was done on a second by second basis using the Infant and Caregiver Engagement Phases system. The Maternal Sensitivity and Responsivity Scales were used to assess the overall quality of the mother infant interaction.
Hypothesized was that IBAIP would increase positive and sensitive maternal behavior and improve self-regulated infant behavior during mother-infant interaction. Results showed that mothers who received IBAIP were more sensitive in interaction and less overcontrolling towards their infant during the Still-face procedure. Moreover, they showed somewhat more positive behavior during the first interaction episode, which reflects improved baseline interactive behavior. Better regulated infant interaction behavior and less stress behavior was not seen in the intervention infants during the Still-face procedure. We conclude that IBAIP may have led to subtle changes in mother-infant interaction, which are important in view of the developmental improvement of the intervention infants. Small and subtle changes can accumulate and lead to large developmental differences. Further study on the effect of IBAIP on mother-infant interaction should be done to evaluate what part of the intervention procedure may affect improvements in maternal sensitivity that may contribute to the developmentally enhancing effect of IBAIP for preterm infants.

Chapter 3 describes the effect of the IBAIP and very preterm delivery on maternal psychological distress at 6, 12 and 24 months of the infant's corrected age. It was hypothesized that the IBAIP might decrease maternal psychological distress as a result of the support in the home situation provided by professionally trained IBAIP interventionists. Maternal psychological distress was studied using the General Health Questionnaire 28 (GHQ)(Goldberg et al., 1988). At 6 months 125 mothers of preterm babies (64 intervention vs. 61 control) answered the GHQ. Maternal response rates were 86%, 76% and 62% at 6, 12 and 24 months,

respectively. No differences in maternal psychological well being were found between mothers of infants who received IBAIP after discharge from the Neonatal Intensive Care Unit, compared those who did not. In general, all mothers reported much psychological stress at 6 months after term date, as more than half (56%) of the mothers then scored above the clinical cut-off level. Because of the high amount of mothers with clinically relevant psychological distress symptoms at 6 months corrected age of their children in both the intervention and control group mothers, we compared our results with mothers of term infants. The significantly higher rate of clinical cases (35%) in the group mothers of very preterm infants showed that mothering a term infant, is not as distressing as mothering a very preterm infant. However, two years after birth the psychological stress levels in both the intervention and control group mothers of preterm infants have normalized compared to norm reference women. More focused intervention for the mother is warranted during the first 6 months after preterm delivery.

Chapter 4 focuses on the effect of the IBAIP on maternal parenting stress 12 and 24 months after preterm delivery. It was hypothesized that the IBAIP might decrease maternal parenting stress as a result of the support at home by the professional interventionists. Maternal parenting stress was assessed with the Dutch version of the Parenting Stress Index (NOSI). At 12 months 123 mothers completed the NOSIK (response rate 79%) and at 24 months 103 mothers completed the NOSI (response rate 67%). Mothers who received IBAIP assessed their children at 24 months corrected age as happier and less hyperactive/distractible than the control mothers. However, the intervention group mothers reported at the same time more feelings of social isolation. More information from the parents is needed to detect the aspects of the IBAIP which may have contributed to these feelings of social isolation. The IBAIP interventionist should become aware of this possible consequence of the intervention.
No differences in maternal parenting stress between the intervention and control group were found at 12 months. Weak to moderate correlations were found between maternal parenting stress and maternal psychological distress at 12 and 24 months. Parenting a very preterm infant, even a very high risk infant, was not found to lead to extreme levels of parenting stress 1-2 years after birth.

In **Chapter 5** the effect of the IBAIP on maternal attachment representations is studied. It was hypothesized that IBAIP support might have a positive impact on the mother-child relationship, reflected in the maternal

attachment representations regarding her child and her relationship with her child. Maternal attachment representations were assessed with the Working Model of the Child Interview. This interview is designed to elicit parents' feelings about their relationships with their children. It was also used in order to allow mothers to express themselves in their own words and to emphasize, what they remembered as important feelings and experiences. Next to the resulting attachment classifications, conventional content analysis was used to gain a richer understanding of maternal emotions and experiences after very preterm delivery.
Seventy-eight mother-infant dyads participated in the interview. No differences were found in the quality of maternal representations of the attachment relationship with their child in the intervention and control group mothers. In both groups about 70% of the mothers had a balanced attachment representation and 30% had a nonbalanced attachment representation of their relationship with their infant.
Content analyses showed that 50% of the mothers felt negative emotional responses to their infant when they first saw their baby. These negative emotions consisted of feelings of anxiety and alienation. The first few weeks at home with the child after discharge were described as stressful, worrisome and anxious by 39% of the mothers. Negative emotions during these first moments with their baby were related to a nonbalanced maternal attachment representation. Therefore, it seems very important to support mothers both early during their infants' stay in the hospital, as well as later at home after hospital discharge, to build a healthy attachment relationship.

In **Chapter 6** the results of the study are discussed, followed by a critical review of the strengths and limitations of the study, the clinical implications and considerations for future research. It is concluded that the IBAIP led to more sensitive mother-infant interaction at 6 months after term date and to some decreases in maternal parenting stress at 24 months after term date. However, the IBAIP in its current form did not prevent high levels of maternal psychological distress during the first year after birth and may have caused feelings of isolation two years after term date. Further study is necessary to detect which specific aspects of the IBAIP are responsible for these results. Implementation of the IBAIP is justified, but the current intervention program needs to be extended with specific maternal support to prevent clinical levels of psychological distress and to promote a secure mother-infant bonding.

Samenvatting

(Summary in Dutch

Samenvatting

Jaarlijks wordt 8% van alle kinderen in Nederland ernstig prematuur geboren. Ernstige prematuriteit heeft consequenties voor zowel het kind als de ouder. Premature kinderen lopen een verhoogd risico op neurologische beschadigingen in de kindertijd ten gevolge van neonatale hersenbeschadiging alsmede door een onderbreking van de normale hersenrijping die plaatsvindt tijdens het laatste trimester van de zwangerschap. Naast en vaak door medische complicaties hebben premature kinderen problemen met zelf-regulatie, dat wil zeggen het vermogen om emoties af te stemmen, zich zelf te kalmeren, bevrediging uit te stellen en verandering in de omgeving te tolereren. Dit beïnvloedt hun mogelijkheden om te exploreren en te interacteren met hun omgeving en derhalve hun leerprocessen.
Voor ouders onderbreekt vroeggeboorte het normale aanpassingsproces van het ouderschap, niet alleen vanwege de plotselinge geboorte, maar ook vanwege de kwetsbare medische conditie van het kind en de extra lange ziekenhuisopname. Het omgaan met een vroeggeboorte kan een moeilijke en stressvolle ervaring zijn voor ouders en kan leiden tot posttraumatische stress symptomen. Premature geboorte kan ook de ontwikkeling van een gezonde moeder-kind relatie bemoeilijken. Zowel de NICU omgeving, waarin fysieke nabijheid en verzorging van het kind door de moeder niet vanzelfsprekend zijn, als de emotionele disbalans van de moeder maakt het haar moeilijk om zich te hechten aan haar kind. Ook de moeder-kind interactie na vroeggeboorte kan lastiger zijn door de onduidelijke gedragssignalen van de baby en de emotionele staat van de moeder die haar mogelijkheden om sensitief om te gaan met haar kind beïnvloedt. Aangezien de ontwikkelings-, sociaal-emotionele en gezondheidsproblemen van zeer prematuur geboren kinderen zich zelfs tot in de vroege volwassenheid kunnen voordoen, hebben allerlei type vroege interventie programma's geprobeerd om de lange termijn uitkomsten van deze kinderen te verbeteren. Tot nu toe zijn door slechts een aantal interventie programma's na ontslag alleen lange termijn effecten gevonden op de cognitieve ontwikkeling. Maar effecten van zulke programma's op de emotionele en functionele staat van de ouders zijn zelden onderzocht.

In dit proefschrift zijn de effecten van het Infant Behavioral Assessment en Interventie Programma (IBAIP) geëvalueerd. IBAIP is een preventief interventie programma na ontslag uit het ziekenhuis en is gericht op ondersteuning van de zelf-regulatie van het kind als een van de basis

elementen die ten grondslag liggen aan de ontwikkeling van het kind en de ouder-kind relatie en interactie. **(Hoofdstuk 1)**
De effecten van de IBAIP zijn onderzocht in een gerandomiseerde trial in een groep ernstig prematuur geboren kinderen en hun ouders. Focus was de moeder-kind interactie, het psychisch welbevinden van de moeder, opvoedkundige stress van de moeder en de hechtingsideeën van de moeder.

In **Hoofdstuk 2** is het effect van de IBAIP op de moeder-kind interactie beschreven. Moeder-kind interactie werd geobserveerd tijdens de Still-face procedure waaraan 112 moeder-kind paren deelnamen, 57 uit de interventiegroep en 55 uit de controlegroep. De Still-face procedure bestaat uit drie episodes. In de eerste 2-minuten durende episode mag de moeder met haar kind spelen zoals ze dat thuis ook doet. Tijdens de 2-minuten durende Still-face episode wordt de moeder geïnstrueerd om geen contact te maken met haar kind en met een uitdrukkingsloos gezicht langs haar kind heen te kijken. Tijdens de derde episode mag de moeder weer contact maken met haar kind. Micro-analytische codering (per seconde) aan de hand van het Infant and Caregiver Engagement Phases systeem werd gebruikt. De Moederlijke Sensitiviteit en Responsiviteit Schalen werden gebruikt om de algehele kwaliteit van de moeder-kind interactie te beoordelen.
De hypothese was dat de IBAIP positief en sensitief interactiegedrag van de moeder zou bevorderen en zelf-regulerend gedrag van het kind zou verbeteren. De resultaten laten zien dat moeders die IBAIP hebben gehad sensitiever en minder directief waren ten opzichte van hun kind tijdens de Still-face procedure. Bovendien lieten ze ook meer positief interactiegedrag zien tijdens de eerste interactie episode, wat mogelijk een weerspiegeling is van verbeterd baseline interactiegedrag.
Beter regulatiegedrag bij het kind en minder stress gedragingen werden niet gevonden in de interventie kinderen tijdens de Still-face procedure. We concluderen dat IBAIP mogelijk tot subtiele veranderingen in de moeder-kind interactie heeft geleid, die belangrijk zijn met het oog op de betere ontwikkelingsuitkomsten van de interventie kinderen. Kleine en subtiele veranderingen kunnen zich opeenstapelen en tot grote verschillen in ontwikkeling leiden. Vervolgonderzoek naar het effect van de IBAIP op moeder-kind interactie is nodig om er achter te komen welk onderdeel van de interventie procedure verantwoordelijk is voor de verbeteringen in moederlijke sensitiviteit en die mogelijkerwijs bijdragen aan de betere ontwikkelingsuitkomsten ten gevolge van de IBAIP voor premature kinderen.

Hoofdstuk 3 beschrijft het effect van de IBAIP en premature geboorte op psychische stress van de moeder op 6, 12 en 24 maanden (gecorrigeerd voor vroeggeboorte). De hypothese was dat IBAIP moederlijke psychische stress zou verminderen door de ondersteuning thuis die gegeven werd door IBAIP interventionisten. Psychische stress van de moeder werd gemeten met de General Health Questionnaire 28 (GHQ). Op 6 maanden vulden 125 moeders van prematuur geboren kinderen (64 interventie vs. 61 controle) de GHQ in. De responspercentages van de moeders waren achtereenvolgens 86%, 76% en 62% op 6, 12 en 24 maanden. Er werden geen verschillen gevonden in het psychisch welbevinden van de moeders van kinderen die IBAIP na ontslag uit het ziekenhuis hadden gehad, en de moeders die geen IBAIP hadden gehad. Over het algemeen gaven alle moeders 6 maanden na de geboorte van hun kind (gecorrigeerd voor vroeggeboorte) veel psychische stress aan, aangezien meer dan de helft (56%) van de moeders in de klinische range scoorde. Omdat in zowel de interventie- als de controlegroep zoveel moeders klinisch relevante psychische stress rapporteerden op dit tijdstip, hebben we onze resultaten vergeleken met een groep moeders van op tijd geboren kinderen. Het significant hogere aantal klinische scores (35%) in de groep moeders van prematuur geboren kinderen liet zien dat moeder zijn van een op tijd geboren kind niet zo stressvol is als van een prematuur geboren kind. Maar 2 jaar na de geboorte van hun kind is de psychische stress zowel in de interventie- als in de controlegroep moeders genormaliseerd en vergelijkbaar met de norm. Specifieke interventie die aandacht besteedt aan de impact van ernstige vroeggeboorte op het psychisch welbevinden van de moeder wordt aanbevolen tijdens de eerste 6 maanden na de bevalling.

Hoofdstuk 4 focust op het effect van de IBAIP op de opvoedkundige stress van de moeder 12 en 24 maanden na de premature geboorte. De hypothese was dat de IBAIP de moederlijke opvoedkundige stress zou kunnen verminderen door de thuisondersteuning gegeven door IBAIP interventionisten. Opvoedkundige stress van de moeder was gemeten met de Nijmeegse Ouderlijke Stress Index (NOSI). Op 12 maanden vulden 123 moeders de NOSIK in (responspercentage 79%) en op 24 maanden vulden 103 moeders de NOSI in (responspercentage 67%). Moeders die IBAIP hadden gehad beoordeelden hun kinderen als vrolijker en minder hyperactief/afgeleid dan de controlegroep moeders. Maar de interventiegroep moeders rapporteerden tegelijkertijd meer gevoelens van sociale isolatie. Meer informatie is nodig van de ouders om te achterhalen welke aspecten van de IBAIP mogelijkerwijs hebben bijgedragen aan

deze gevoelens van sociale isolatie. De IBAIP interventionisten moeten zich realiseren dat dit een mogelijke consequentie is van de interventie.
Er werden geen verschillen in opvoedkundige stress van de moeders gevonden op 12 maanden. Zwakke tot middelgrote correlaties werden gevonden tussen de opvoedkundige stress van de moeders en de psychische stress op 12 en 24 maanden. Ouder zijn van een ernstig prematuur geboren kind, zelfs een zeer hoog risico kind, leidt volgens de resultaten van deze studie niet tot extreme niveaus van opvoedkundige stress 1-2 jaar na de geboorte.

In **Hoofdstuk 5** is het effect van de IBAIP onderzocht op de hechtingsrepresentaties van de moeder. De hypothese was dat IBAIP ondersteuning een positief effect zou kunnen hebben op de moeder-kind relatie, weergegeven in de hechtingsideeën van de moeder ten opzichte van haar kind en haar relatie met haar kind. Moederlijke hechtingsideeën werden gemeten met het Working Model of the Child Interview. Dit interview is ontwikkeld om de gevoelens van ouders over hun kinderen in kaart te brengen. Het interview werd ook gebruikt om moeders de kans en ruimte te geven om zich in hun eigen woorden te kunnen uitdrukken en te benadrukken wat zij als belangrijkste gevoelens en ervaringen konden herinneren. Niet alleen de hechtingskwalificatie werd gebruikt om een beter inzicht te krijgen in de moederlijke emoties en ervaringen na een zeer premature geboorte, maar ook een conventionele inhoudsanalyse.
Achtenzeventig moeders namen deel aan het interview. Er werden geen verschillen gevonden in de kwaliteit van de moederlijke hechtingsideeën tussen de interventie- en controlegroep moeders. In beide groepen had ongeveer 70% van de moeders een evenwichtige (balanced) hechtingsrepresentatie en 30% had een onevenwichtige (nonbalanced) hechtingsrepresentatie van hun relatie met hun kind.
Inhoudsanalyses lieten zien dat 50% van de moeders negatieve emotionele reacties vertoonde bij het voor de eerste keer zien van hun kind. Deze negatieve emoties bestonden uit gevoelens van angst en vervreemding. De eerste weken thuis met het kind na ontslag uit het ziekenhuis werden door 39% van de moeders als stressvol, zorgelijk en angstig beschreven. Negatieve emoties tijdens deze eerste momenten met hun baby waren gerelateerd aan een onevenwichtige (nonbalanced) hechtingsrepresentatie. Het lijkt daarom erg belangrijk om moeders te ondersteunen zowel tijdens de opname van het kind als later in de thuissituatie na ontslag uit het ziekenhuis, om een gezonde hechtingsrelatie op te bouwen.

In **Hoofdstuk 6** worden de resultaten bediscussieerd, gevolgd door een kritische review van de sterke en zwakke punten van de studie, de klinische implicaties en overwegingen voor toekomstig onderzoek. Geconcludeerd wordt dat de IBAIP tot meer sensitieve moeder-kind interactie leidt 6 maanden na de uitgerekende datum en tot een afname van bepaalde aspecten van opvoedkundige stress 24 maanden na de uitgerekende datum. Maar de IBAIP in zijn huidige vorm heeft niet de hoge niveaus van psychische stress in moeders tijdens het eerste jaar na de geboorte kunnen voorkomen en kan gevoelens van sociale isolatie hebben veroorzaakt 2 jaar na de uitgerekende datum. Vervolgonderzoek is nodig om uit te vinden welke aspecten van de IBAIP verantwoordelijk zijn voor deze resultaten. Implementatie van de IBAIP is gerechtvaardigd, maar het huidige interventieprogramma dient te worden uitgebreid met specifieke ondersteuning voor de moeder om klinische niveaus van psychische stress te voorkomen en een veilige moeder-kind hechting te bevorderen.

Appendix I

List of abbreviations

APPENDIX I ABBREVIATIONS

AAI	Adult Attachment Interview
BSID-II-NL	Bayleys Scales of Infant Development (Dutch version)
CBCL	Child Behavior Check List
GHQ	General Health Questionnaire
IBA	Infant Behavioral Assessment
IBAIP	Infant Behavioral Assessment and Intervention Program
ICEP	Infant and Caregiver Engagement Phases
IHDP	Infant Health and Development Program
IVH	Intraventricular Hemorrhage
MITP	Mother Infant Transaction Program
MSRS	Maternal Sensitivity and Responsivity Scales
NICU	Neonatal Intensive Care Unit
NIDCAP	Newborn Individualized Developmental Care and Assessment Program
NOSI(K)	Nijmeegse Ouderlijke Stress Index (Kort)
PSI	Parenting Stress Index
PTSD	Post Traumatic Stress Disorder
PVL	Periventricular leucomalacia
RCT	Randomized Controlled Trial
SF	Still-Face
SGA	Small for Gestational Age
VLBW	Very Low Birth Weight
WMCI	Working Model of the Child Interview

Appendix II

Parent quotations in English

APPENDIX II PARENT QUOTATIONS IN ENGLISH

Page 7:
The only thing I can remember about giving birth, is that it had to happen all of a sudden, although I had already been in the hospital for three weeks. I was put under an anaesthetic. When I woke up, my boyfriend came to me and said: "We have a son." I thought: who are you, a son? It wasn't until the next day that I was brought to see T. The word delivery does not bring up any feelings. I was sick, I needed surgery, there was this patient in the hospital and this patient was my son. While I was still lying in my bed, they brought me to the NICU, and then drove me to one of those small glass houses and told me that the little one inside was my son. I remember thinking: it just as well could have been any of the other houses. All kinds of scenarios went through my mind, of those scary movies in which children get switched.

Page 27:
After 2,5 months he came home, and I had been looking forward to that so much, to finally be together. But it was not like that at all, because he was crying all the time. I tried everything, and I really mean everything, because I felt like I had to make up for lost time, but it was as if nothing was good enough...

Page 53:
After T. came home and he was doing okay, I broke down. Of course I could have asked for help, but I felt that we had to make up for lost time. At least six months I kept going, although I was actually feeling ill. I lost myself for a while. I hyperventilated and had to stay home from work. Nobody understood. It wasn't like I needed consolation from everyone, but some sympathy would have been nice.

Page 69:
After every single test I had to hear that he wasn't doing well, that he was developmentally behind, while I was so proud that he had rolled over in his bed. He is much more special and brighter than other children. He has come through everything and survived. I have a lot of respect for that. Well, respect is not the right word. In the hospital they've always said he is a fighter, and it sort of made me laugh, but it's really the truth.

Page 87:
I have been worried about our attachment. I always felt some distance. Not consciously, but I always had that idea in the back of my mind that he could still die or become very ill... However, I knew we were okay when one day I dropped a knife in the kitchen and I stretched my arm to catch it before it could hurt my son. But that happened more than a year after his birth.

Page 107:
I have experienced talking to our IBAIP-interventionist as something very positive, and I still do. When I felt insecure about how to handle my baby, she was able to give me support and keep me on the right track. She also gave me the feeling that, as a parent, I know what is the right thing to do and I have everything I need to help my baby. She has helped me become much more self-confident, and she taught me to watch my baby and see what works best for him.

Appendix II

Infant Behavioral Assessmen

APPENDIX III INFANT BEHAVIORAL ASSESSMENT

Observer: _____ Child: _____
Birthdate: _____
Gestational Age: _____

Observation #: 1 2 3 4
Dates: / / / /

AUTONOMIC / VISCERAL									MOTOR								STATE							
Color	Pink								**Arms**	Reach								Active Alert						
	Mottled									Well-Regulated Tone								Hyperalert						
	Pale									Smooth Movement								Cry						
	Red									Arm Over Face								ATTENTION / INTERACTION						
	Dusky									ATNR								**Eyes**	Facing Gaze					
Respiration	*Stable									Stop									Directed Gaze					
	Yawn									Bow									Brow Raising					
	Sigh									Airplane									Animate Locking					
	Irregular									Flaccid									Inanimate Locking					
	Sneeze									Straighten w/Tension									Hand Gaze					
	Cough									Shoulder Retraction									Gaze Aversion					
	Hiccough								**Hands**	Grasp									Brow Lowering					
	Gasp									Resting									Blink					
	Pause									Holding On									Clench					
Visceral	*Stable									Hand to Midline									Upward Gaze					
	Burp									Hand to Mouth								**Expression**	Smile					
	Spit Up									Groping									Ooh Face					
	BM Grunt									Hand on Stomach									Facial Brightening					
	Gag									Self-Clasp									Sober					
	Elimination									Hand on Head									Lip Compression					
	Vomit									Finger Extension									Wary					
Neurophysiological	*Stable									Finger Splay									Frown					
	Tremor									Fisting									Pout					
	Twitch								**Legs**	Well-Regulated Tone									Grimace					
	Startle									Smooth Movement									Ugh Face					
	Seizure									Bracing									Gape Face					
MOTOR										Toe Grasp									Cry Face					
Head	Orients									Foot Clasp								**Oral**	*Neutral					
	Lowering									Toe Splay									Sucking					
	Headshake									Flaccid									Mouthing					
	Maximal Head Turn									Sitting on Air									Tongue Show					
Trunk/Extremities										Straighten w/Tension									Suck Search					
	Well-Regulated Tone								STATE										Drooling					
	Stilling									Deep Sleep									Tongue Extension					
	Tuck									Light Sleep									Jaw Extension					
	Immobility									Drowsy								**Vocal**						
	Squirm									Diffuse Alert									Pleasurable					
	Pull Away									Alert									Undifferentiated					
	Flaccid									Interactive Alert									Protest					
	Arching																							

Adapted from Als, 1984

Appendix IV

Evaluation form parent

APPENDIX IV EVALUATION FORM PARENTS

STIPP

Evaluatie STIPP thuisbegeleiding van: , inclusienr.
Thuisbegeleidster:

STIPP thuisbegeleiding	**Altijd / Volledig**	**Grotendeels wel**	**Deels wel , deels niet**	**Grotendeels niet**	**In het geheel niet**
Waren de ouders bereid rekening te houden met de STIPP afspraken?					
Heb je alle 8 STIPP sessies gebruikt?					
Was het mogelijk een vertrouwensbasis te bewerkstelligen met de ouders?					
Hebben de STIPP adviezen de ouders bevestigd in de omgang met hun kind?					
Hebben de STIPP adviezen de ouders ook nieuwe informatie of tips gegeven over het omgaan met uw kind?					
Waren de ouders in staat de informatie en tips zelfstandig te gebruiken in de omgang met uw kind?					
STIPP verslagen voor ouders					
Is het je gelukt na elke sessie een STIPP verslag te schrijven voor de ouders?					
Werden de STIPP- verslagen gelezen door de ouders?					
Heb je de indruk dat de STIPP verslagen door de ouders werden gebruikt in de omgang met hun kind?					
Werden de STIPP- verslagen ook gebruikt ter informatie van familie, vrienden of oppas van het kind?					
Heb je gebruik gemaakt van andere middelen dan STIPP verslagen? Zo ja, welke:					
Duur en frequentie van de thuisbegeleiding	**Onvoldoende**	**Nogal kort/ nogal weinig**	**Voldoende**	**Ruim Voldoende**	**Te veel/ Te lang**
Wat vond je van de tijdsduur van een STIPP huisbezoek?					
Wat vond je van het aantal STIPP huisbezoeken in 6 maanden?					
Evaluatie STIPP	**Ja, zeker**	**Ja, een beetje**	**Neutraal**	**Matig**	**Nee**
Heb je positieve effecten gezien bij de baby als gevolg van de tips en informatie die je gegeven hebt?					
Heeft de STIPP thuisbegeleiding naar jouw mening een gunstige invloed gehad op het contact tussen de ouders en hun baby?					
Denk je dat je de ouders hebt kunnen steunen met STIPP thuisbegeleiding?					
Denk je dat adviezen ook invloed hebben gehad op de omgeving en/of levensstijl van de ouders?					
Vind je dat dit kindje voortzetting van de begeleiding nodig heeft?					
Vind je dat deze ouders voortzetting van de begeleiding nodig hebben?					
Heb je bij deze ouders de STIPP thuisbegeleiding als plezierig ervaren?					
Vond je voor deze ouders en hun kind de thuisbegeleiding de investering waard?					

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Appendix V

Acknowledgements

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Appendix VI

About the author

APPENDIX VI ABOUT THE AUTHOR

Dominique Meijssen was born on November 7th, 1978 in Alkmaar. In 1997 she received her Gymnasium diploma at the Murmellius Gymnasium in Alkmaar. Because she didn't exactly know what to do next, she moved to Spain to study Spanish. Back in the Netherlands she studied German literature at the University of Amsterdam and after obtaining her propaedeutic degree she switched in 1999 to Pedagogical sciences. She wrote her MA thesis on the experiences and need for support in parents of very preterm infants after hospital discharge from the Leiden University Medical Center. In 2004 she started her PhD project at the Department of Pediatric Psychology at Tilburg University in association with the Academic Medical Centre (AMC) in Amsterdam. In 2006 she started working at the neonatal follow up in the AMC neonatal outpatient clinic next to her research activities. Since 2009 she participates in a research project in the Sint Lucas Andreas hospital (SLAZ) on specialized care for children from different ethnic backgrounds with chronic diseases. Besides her SLAZ activities, Dominique is currently working as a researcher in the AMC as part of the IBAIP implementation project and as a psychologist at Perspectief Advies, a primary health care practice in Amsterdam.